Parkinson's disease in primary care

A joint journey of patients and general practitioners

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Preface

Mrs. P.D., a seventy-year-old who had always enjoyed good health, visits her GP with defecation problems. She has suffered from constipation for quite some time, but blood tests, an ultrasound scan and a colonoscopy in the past year showed no abnormalities. The GP, therefore, has prescribed her laxatives for a number of weeks now, and these appear to be helping her.

Well over a year after that, Mrs. P.D. revisits her GP. She mentions that her right shoulder is troubling her considerably. The past few months, she has had to cancel tennis appointments more and more often because she cannot manage to throw the ball for her service. Carrying a heavy shopping bag is also beyond her possibilities. Her energetic spouse encouraged her to go and see her GP. After a physical examination, showing clear indications of a frozen shoulder, the GP decides to refer Mrs. P.D. to an orthopedic surgeon, who treats her with corticosteroid injections several times. These appear to be providing some alleviation.

Another year later, Mrs. P.D. visits her GP again, this time because she feels down and has difficulty concentrating. At home she cannot be bothered to do anything. Her housekeeping is a burden to her. She has lost a lot of weight for lack of appetite. She has quit her tennis because it was all taking too much out of her. Her spouse has gradually taken over more and more of her tasks: he does the cooking and the shopping as his wife tends to forget more and more things on the list. Suspecting her of having a depression, the GP consults a psychiatrist. The psychiatrist finds Mrs. P.D. absent-minded and confused and suspects her of being in the early stages of dementia.

Six months later, Mrs. P.D. visits the general practice again with her spouse. While she walks from the waiting room to the consulting room, the GP notices that she barely lifts her feet and walks hunched over. Besides her old complaints of constipation, stiffness in the right shoulder, and gloominess, Mrs. P.D. is trembling increasingly, to the point where her old tennis friends were wondering whether she was nervous. Things are also getting tougher on her spouse. The GP has the feeling there is something more going on. The physical examination shows rigidity and tremor on the right-hand side. The GP explains to Mrs. P.D. and her spouse that he is thinking of Parkinson's disease and proposes referring her to a neurologist.

After her visit to the neurologist and almost three years after her initial complaints started, the GP's suspicion is confirmed: Mrs. P.D. has Parkinson's disease.

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General introduction

Background

Exactly 200 years ago, in 1817, the British surgeon James Parkinson wrote his *Essay on the Shaking Palsy* and defined it as 'Involuntary tremulous motion, with lessened muscular power, in parts not in action and even when supported; with a propensity to bend the trunk forwards, and to pass from a walking to a running pace: the senses and intellects being uninjured'.[1] At that time, his essay drew little attention. Half a century later, the French neurologist Jean-Martin Charcot acknowledged Parkinson's observations and named the disorder that he believed was characterized by bradykinesia, tremor at rest, rigidity, facial immobility and disturbances of gait and posture *maladie de Parkinson*.[2] It took until the 20th century before degeneration of dopamine-producing cells in the substantia nigra of the brain was recognized as the cause of the disease and pharmacological treatment for Parkinson's disease (PD) was introduced.[3]

Although the diagnosis of PD still requires the clinical presence of bradykinesia and muscular rigidity, a 4-6 Hz resting tremor or postural instability, our understanding of the symptom complex of PD has grown since the 1800s.[4] Nowadays, PD is recognized as a multisystem disorder consisting of both motor and non-motor symptoms.[5] The non-motor symptoms vary from autonomic disturbance (i.e. orthostatic hypotension, constipation, urinary incontinence and sexual dysfunction) to sleep disorders, pain, olfactory dysfunction and neuropsychiatric disorders such as depression, anxiety, hallucinations and cognitive impairment.[6] The clinical expression of the motor and non-motor symptoms, in addition, varies between patients, and patients' perceptions of the most troublesome symptoms differ as well.[7-9] It is because of this complexity that a multidisciplinary approach to care, with the patient's active participation, is recommended.[10]

Above all, there is one health care provider involved in the care of PD patients from the first symptom to advanced-stage PD: the general practitioner (GP). Therefore, this thesis focuses on the role of primary care in Parkinson's disease.

Towards the diagnosis of Parkinson's disease

PD is known to have an insidious onset. Years before patients are being diagnosed, they may already be suffering from symptoms related to the disease.[11, 12] Nevertheless, many hurdles have to be cleared to get from the first symptom to the moment of diagnosis. Patients, for example, do not always recognize that their symptoms require medical attention, and consultation of a health care provider does not guarantee an instant diagnosis.

Patients

Bodily changes do not immediately lead to symptoms. Patients first need to pay attention to and become aware of physical and mental sensations. As a next step, the sensations they experience need to be interpreted as symptoms that must be evaluated for their perceived health threat.[13, 14] The representations patients make of their illness determine their coping

strategies, including the decision to seek medical attention and the actual act of doing so.[15] Breen et al. already found that PD patients need time to recognize their motor symptoms and to realize that medical attention is required.[16] In addition, there are substantial differences between individuals in frequency of and reasons for contacting their GP, irrespective of PD signs and symptoms.[17] Research in patients with cancer and diabetes mellitus has shown that a number of factors influence the pathway from the first recognizable symptom to the diagnosis. These factors include the nature of the symptoms and the patients' emotional response to them, knowledge of the disease, the consultation of significant others and concurrent problems in patients' personal lives.[18, 19] Knowledge of the factors influencing the course of the diagnostic pathway of PD, however, is scarce.

Patients' experiences during the diagnostic process do not only influence their feelings about the pathway itself. Studies amongst cancer patients found that patients tend to lose confidence and trust in their GP if they need to pay multiple visits to the general practice before being referred.[20-22] Patients may interpret the need for repeatedly consulting their GP as lack of responsiveness and, therefore, as diagnostic delay, resulting in dissatisfaction and worse long-term care experiences.[23] In addition, patients suffering from cancer types that are difficult to recognize, such as ovarian cancer, are known to change over to another general practice more often than patients with cancer types that are easier to recognize.[24] PD may be difficult to recognize as well, and the diagnostic pathway can be lengthy and uncertain. It is reasonable, therefore, to expect a negative influence of patients' diagnostic experiences on their satisfaction, confidence and trust and, hence, on the patient-doctor relationship.[16, 25] Nonetheless, we do not know how PD patients evaluate their diagnostic pathway.

General practitioners

Symptoms already present before the onset of the typical motor signs of PD are known as prodromal symptoms.[11] Prodromal symptoms include, for example, shoulder pain and restrictions in movement, olfactorary dysfunction and rapid eye movement sleep behavior disorders.[5, 12] Most prodromal symptoms of PD are not disease-specific.[12] In the Netherlands, patients generally first present their symptoms to their GP. As the gatekeepers to specialist care, GPs need to discriminate the prodromal symptoms of PD from symptoms related to other causes and subsequently decide on referral.[26, 27] Although PD is the second most common neurodegenerative disorder after Alzheimer's disease, it is rather uncommon in general practice in the Netherlands with an incidence varying from 0.3 per 1,000 person-years in subjects aged 55-65 years to 4.4 per 1,000 person-years for subjects aged ≥85 years and a prevalence of 1.3%.[28, 29]

Recognizing PD as the cause of the presented prodromal symptoms, therefore, can be challenging.[25] Adding to the diagnostic difficulties is the fact that earlier studies on prodromal symptoms are mainly hospital based and focus on patients referred to neurologists. When patients consult their GP, symptoms are often still limited and embedded in clinical uncertainty, while referral to the neurologist takes place at a later point in the disease trajectory.[26, 27, 30-33]

As GPs play a crucial role in PD recognition, we need to increase our knowledge of the way prodromal symptoms of PD are presented in general practice.

Changes in care after the diagnosis of Parkinson's disease

Once the diagnosis is set, Parkinson's disease continues to have an influence on the lives of patients and their relatives. Physical, emotional and psychosocial aspects of life are affected by PD.[34, 35] The fluctuating expression of symptoms, which may vary on a daily basis, add to the impact of the disease.[34, 35] Progression of PD, in addition, will confront patients and their relatives with new disabilities and limitations.[36] In some cases, the support or medical care that is offered to patients needs to be changed to cope with these disabilities and limitations. As 80% off all PD patients in the Netherlands continue to live at home during the course of their disease, it is likely that GPs can play a role in offering support during these changes in care.[37]

Patients

PD patients are likely to experience a variety of changes in care. So far, research on changes in care has mainly focused on transitional care from one health care setting to another or to home.[38-42] Such changes jeopardize continuity of care and challenge both the patients' and the health care providers' communication skills as there is a need for clarity in preferences, expectations and roles in care.[40, 43] Studies among patients with prevalent chronic conditions such as heart failure, chronic obstructive pulmonary disease (COPD) and cancer show that patients feel they lack control when being discharged from hospital to home. In addition, they feel uncertain, unprepared, inadequately guided and not involved in care-taking decisions.[41, 42] Changes in care may also be more closely related to patients' personal context, such as changes in the help they receive from a relative or adjustments made to their living situation. Although these changes might be experienced without the direct involvement of health care providers, patients may still benefit from support. For this reason, we wonder what changes in care PD patients actually experience and how they cope with them, yet information on this topic is limited.

General practitioners

GPs are important health care providers to offer support during changes in care. In the Netherlands, all citizens are registered with a general practice.[44] This structure supports Dutch GPs in their role as family doctors, who are informed about the physical and mental state of all family members and the contextual circumstances that influence their well-being. Amongst other tasks, GPs play an important role in disease-specific care for prevalent chronic conditions such as COPD and diabetes.[45, 46] Knowledge of their patients' personal context facilitates and enhances the quality of such care.[47] The long-term patient-doctor relationship, in addition, facilitates the GPs' understanding of their patients' ability to cope. The GPs' knowledge of their patients' contextual circumstances enables them to offer personalized support during changes in care.

However, for a complex chronic condition such as PD - a condition with a versatile presentation and a complicated treatment regimen - disease-specific care is usually best offered by specialized health care providers with expert knowledge and skills.[36, 47] Although GPs are aware of the impact the disease and possible changes in care may have on the patients' context, they are not involved in disease-specific care. Nevertheless, the GPs' experience in offering support during changes in care may be valuable for PD patients as well. In order to know if GPs can offer this support to PD patients, we need to increase our understanding of the role of GPs in PD care.

Rationale of this thesis

Specialized health care providers now agree that PD patients benefit from early intervention and treatment, taking into account the patients' personal situation and care preferences and discussing the benefits of intervention on the one hand and the risk of side effects on the other. Research has shown that early intervention is likely to result in maintenance of quality of life, slowdown of clinical progression and reduction of mortality.[48-50] However, before interventions can be discussed, the disease has to be diagnosed. In order to allow patients and health care providers to take time to discuss referral and treatment steps, delay in the diagnostic pathway should be addressed whenever possible and preferred by patients.

Therefore, we need to understand the factors that influence the course of the experienced diagnostic pathway of PD according to patients. Moreover, a trusting therapeutic relationship between patients and health care providers is indispensable for genuine shared decision-making.[51] As patients and GPs each have a share in establishing such a relationship, we need to improve our understanding of factors that pose a risk to such a relationship by contributing to patients' dissatisfaction with the diagnostic pathway. To facilitate early recognition by GPs, moreover, we need to learn more about the way prodromal PD is presented to GPs.

In addition, for a progressive disease with an extended impact on the lives of patients and their relatives, remarkably little is known about PD patients' (health care) needs during changes in care. Increasing our knowledge of patients' experiences and their expectations of the GP allows for more customized support. In-depth understanding of the difficulties GPs experience in choosing their position in PD care, furthermore, may highlight areas that need attention in order for GPs to provide patient-centered quality care.

Objectives of this thesis

The aim of this thesis is to contribute to patient-centered quality care for community-dwelling patients with PD from the first recognizable symptoms to advanced-stage disease by gaining insight into patients' experiences before, during and after the diagnosis and the challenges GPs are confronted with in providing care to these patients. This thesis, therefore, will address the following objectives:

1. To gain insight into patient experiences of the diagnostic pathway of PD and to describe the factors that influence it.

2. To gain insight into factors influencing patient dissatisfaction with the diagnostic pathway of PD.
3. To characterize the prodromal symptoms of PD presented in general practice.
4. To gain insight into patient experiences and coping with changes in care encountered during the course of PD.
5. To clarify the role community-dwelling PD patients see for their GP in PD care and to clarify the role GPs see for themselves.

Outline of this thesis

Towards the diagnosis of Parkinson's disease

The patient's perspective

Based on a qualitative study of patient essays, ***Chapter 2*** describes the patients' views on the pathway from the first recognizable symptoms to the diagnosis of PD, dividing the pathway into three time intervals: recognizing the symptoms; deciding to seek help; and the process of diagnosing PD.

In ***Chapter 3***, we present data from a quantitative analysis of the above-mentioned patient essays that helps us understand the factors that influence patient dissatisfaction with the diagnostic pathway of PD.

The general practitioner's perspective

Chapter 4 contains a nested case-control study of the prodromal symptoms of PD presented to GPs in four general practices participating in the Continuous Morbidity Registration (CMR) of the Radboud university medical center in Nijmegen, the Netherlands.

Changes in care after the diagnosis of Parkinson's disease

In ***Chapter 5***, we provide the protocol of a longitudinal mixed methods study on changes in care of community-dwelling PD patients using video diaries and in-depth interviews with patients and their GPs.

The patient's perspective

Chapter 6 describes a qualitative analysis of community-dwelling PD patients' experiences and coping with the changes in care they encountered.

The general practitioner's perspective

In ***Chapter 7***, we compare the perspectives of community-dwelling PD patients and their GPs on the role of the GP in PD care, based on the results of a qualitative analysis.

Reflections and recommendations

Chapter 8 provides a critical review of the presented results and the set-up of the studies. In addition, it makes recommendations for clinical practice, education and future research.

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2

Time intervals in diagnosing Parkinson's disease: the patients' views

For me at that time, the image of Parkinson's disease was defined by Prince Claus [member of the Dutch royal family] and the Pope [John Paul II], not knowing they already had an advanced stage of Parkinson's disease. (Female, 47 years)

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Abstract

Objective: To explore patients' views on their pathway to the diagnosis of Parkinson's disease (PD).

Methods: A qualitative study of 52 essays written by patients with PD, using comparative content analysis.

Results: Patients divide their diagnostic pathway into three time intervals: recognition of the symptoms; the decision to seek help; and the process of diagnosing PD. Patients have difficulties recognizing the prodromal symptoms of PD (their knowledge is based on public figures with advanced PD) and mention their general practitioners do as well. The decision to seek help is influenced by the patient's attitude towards health care and by their significant others. More than half of the patients believe their diagnosis was delayed. However, the majority of all patients are satisfied with their diagnostic trajectory.

Conclusion: The pathway to diagnosing PD is an iterative process influenced by patient-, health care provider- and disease-related factors. Despite possible delay in diagnosis, patients are content with their pathway.

Practice implications: In order to facilitate earlier diagnosis and timely therapeutic intervention (in particular with regard to future possibilities for disease-modifying therapy), effort is required to increase the recognition of prodromal symptoms of PD by patients, their significant others and health care providers.

Introduction

Parkinson's disease (PD) is now recognized as a multisystem disorder with motor and non-motor features.[1] Some motor and non-motor features are prodromal symptoms: symptoms that are already present before the onset of the typical motor signs of PD.[2] Patients seem to have prodromal symptoms years before they are diagnosed with PD.[2-4] As physical abnormalities do not immediately lead to symptoms, the process is influenced by attention, awareness, interpretation and attribution of the patient.[5, 6] The illness representations, which patients form of a perceived health threat, influence coping strategies including help-seeking behavior.[7, 8] Earlier research has shown that it takes patients more time to recognize their motor symptoms and to realize they need medical attention, than it takes the general practitioner (GP) to diagnose PD.[9] On the other hand, it is not uncommon that a patient needs to visit a number of health care providers before the diagnosis of PD is made.[10] When it comes to women and patients with young onset PD, health care providers seem to require more time to diagnose PD.[11, 12] However, patients can benefit a lot from an early diagnosis of PD. Early recognition of symptoms allows patients and health care providers to discuss the benefits of timely therapeutic intervention on the one hand and the risk of side effects on the other. They can then make a shared decision on a customized balance between advantages and disadvantages, taking into account the patient's personal situation and preferences. This is likely to result in maintenance of quality of life, slowdown of clinical progression and reduced mortality.[13-15]

Research in patients with cancer has shown that the pathway from the first recognizable symptoms to the diagnosis can be influenced by a number of factors such as the nature of the symptoms and the emotional response to them, knowledge of the disease and the consultation of significant others.[16] In chronic diseases with a less threatening outcome, such as diabetes mellitus, the same factors are of importance.[17] For PD it is unknown which factors influence the diagnostic pathway and how patients reflect on their pathway. However, more insight into the patients' views could lead to interventions that facilitate an earlier diagnosis by avoiding as much delay as possible in the diagnostic pathway. In this study we aim to gain insight into the patients' views on their diagnostic pathway and the factors that influence it. Furthermore, we want to know how patients reflect on their pathway.

Methods

Recruitment

This study is part of a larger study on the prodromal symptoms of PD and the patients' views on the trajectory towards the diagnosis. For this reason, all patient members of the Dutch Parkinson's Disease Association whose email addresses were known (n = 4717) received an email, in which the study was explained and they were asked to participate. In case patients were willing to participate a digital essay format was sent. Digital essays rather than individual interviews were chosen to assure easily accessible, anonymous participation and to enable patients to reflect in their own pace. Participation was voluntarily and one-time only.

Patients were provided with contact information in case of questions, concerns or hesitations about participation. After completion of the essay format, patients had to agree with submission of the format. This step was assessed as informed consent.

Data collection

Patients were asked for their demographic characteristics at the time of diagnosis: sex, age, level of education, employment status and civil status. To help them formulate their essay, a number of questions, based on literature and expert opinion, were developed in a small pilot study. The final questions are shown in Table 1.

	Question
1.	Can you describe your first complaint(s) that eventually turned out to be a forerunner sign of PD? What did you do when you experienced this/these complaint(s)?
2.	Can you describe what happened next, until the moment you were diagnosed with PD?
3.	Did people in your surroundings influence the pathway to diagnosis? If so, in what way?
4.	Do you think, in your case, it would have been possible to diagnose PD earlier? If so, at what time and why do you think so? What were the consequences for you and your family?

Table 1. Subject questions included in digital essay format

Of all the patients who received an email, 27% (n = 1251) started writing an essay. Essays were completed by 21% (n = 974) of the patients: 689 patients responded before the reminder, 285 afterwards. Patients with a different diagnosis than idiopathic PD were excluded (n = 74). Finally 900 essays remained (Figure 1).

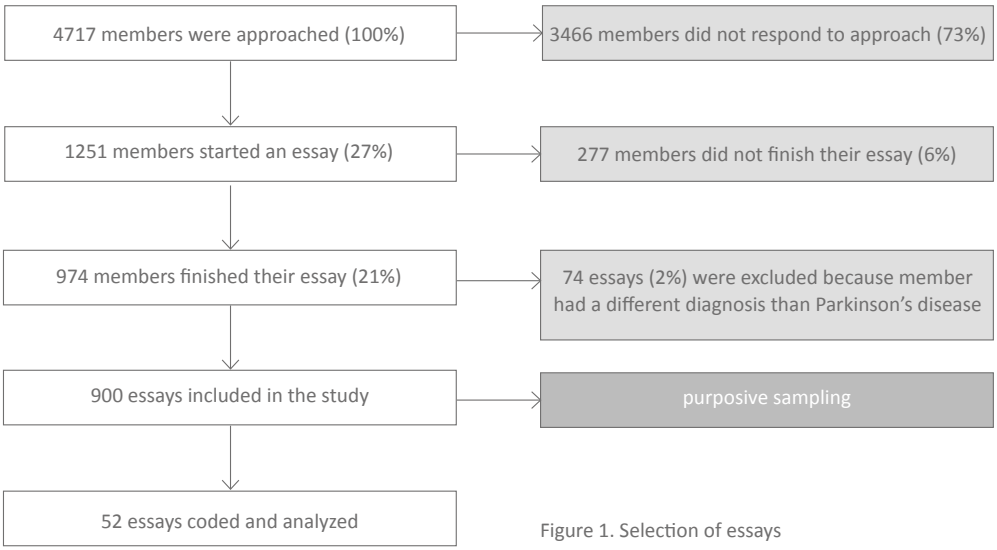

Figure 1. Selection of essays

Data analysis

A purposive sample of 26 essays was entered into ATLAS.ti 7, a software program for detailed coding in qualitative data analysis. Purposive sampling, based on the collected demographic data, was used to increase the external validity and to provide a wide range of patients' experiences. Level of education was divided into two categories: low and high. Finishing elementary school or vocational education was considered a low level of education. Finishing high school, higher professional education or university was considered a high level. The qualitative data analysis was an iterative process by two independent researchers (AP, LB), using the principles of comparative content analysis.[18] The two researchers read the 26 essays several times to familiarize themselves with the data. They independently applied codes to meaningful words and sentences in the essays. These codes were discussed and refined in consensus meetings with the supervisors (ToH, AL). New codes, arising from the discussion, were applied to the essays. After analysis of the 26 essays no significant new codes emerged (saturation). Codes were then grouped into themes, final themes were agreed upon with the supervisors (ToH, AL). These themes were structured in Figure 2. In order to verify the figure and the position of the themes within it, another purposive sample of 26 essays was analyzed.

Time intervals in the pathway to diagnosis of Parkinson's disease		**Recognition of the symptoms**	**Decision to seek help**	**The process of diagnosing PD**
		Iterative process		
Influencing factors on the process	Patient related	Knowledge of PD Alternative explanation for the symptoms Assessment of the severity of the symptoms Influence of family, friends and colleagues	Attitude toward health care Adaptation of lifestyle Influence of the symptoms on daily life Fear for the diagnosis	
	Health care provider related		Doctor-patient communication	Recognition by the health care provider Referral to the neurologist
	Disease related		Alternation of the symptoms	

Figure 2. Stimulating and impeding factors on the pathway to diagnosis of Parkinson's disease (PD); divided into the three time intervals of the pathway to diagnosis and related to factors concerning the patient, the health care provider and the disease

Results

Characteristics of the study population

About half of the participants were women (54%) (Table 2). The mean age at the time of diagnosis was 56 years (SD 14.5), varying between 32 and 84 years. The majority of the patients was employed (62%) and lived with a partner and/or children (75%) at the time of diagnosis.

Demographic variable		n = 52
Sex	Male	24
	Female	28
Mean age in years (SD)	56.3 (14.5)	
Level of education	High	26
	Low	26
Employment	Employee	25
	Self-employed	2
	Retired	12
	Receive sickness benefit	4
	Unemployed	4
	Combination of employments	5
Civil status	With partner/family	35
	Single*	13
	Single* with children	4

*Including widowed and divorced

Table 2 Sample characteristics at the time of diagnosing Parkinson's disease

Diagnostic process

We could distinguish three time intervals in the individual pathways to diagnosis, as described by patients: recognition of the symptoms, the decision to seek help from health care providers and the process of diagnosing PD (Figure 2). Each of these intervals will be further explored beneath.

Recognition of the symptoms

Many patients stressed the importance of recognizing the first symptoms. The majority did not immediately recognize that their symptoms could be part of a disease. Rather, they described they realized something was abnormal. Family, friends or colleagues were described to be of influence in the recognition.

Back from holiday, I thought that the handlebar of my folding bike was loose - it wobbled quite a bit. Perhaps a few days later, I thought the wheel of my regular bicycle wobbled as well. This could not be a coincidence; it had to be me. (Female, 53 years)

[…] I took part in a walking event that spanned four evenings. One of my friends noticed my right arm did not swing while I was walking. (Female, 32 years)

Many patients described they initially found an alternative explanation for their symptoms, sometimes influenced by previous experiences or by family and friends.

I experienced difficulties with writing, which I noticed when writing many Christmas cards in December […]. I blamed increased computer use being the reason I was no longer accustomed to handwriting a lot. (Female, 47 years)

I had some difficulties washing my hair. The right side was slower than the left side. When I said something about it, my children responded: mother you are getting older. (Female, 54 years)

A few patients mentioned that they only knew what advanced PD looked like, based on information from books or television. That was the reason they had not taken PD into consideration.

For me at that time, the image of Parkinson's disease was defined by Prince Claus [member of the Dutch royal family] and the Pope [John Paul II], not knowing they already had an advanced stage of Parkinson's disease. (Female, 47 years)

However, a few patients instantly identified their symptoms as a disease or even as PD.

In February […] my leg started to shake and I knew I had Parkinson's disease (my sister has it too and an acquaintance as well). (Female, 68 years)

Decision to seek help from health care providers

After recognizing that the symptoms were abnormal, patients indicated that they needed to make a decision whether to consult their GP or not. Several patients decided to seek help right after detecting the symptom(s), sometimes stimulated by family, friends or colleagues. However, a few patients seemed to have difficulties assessing the severity of their symptoms. They mentioned they were afraid what the health care provider might think of them and feared that their symptoms were due to dramatizing, or that they were wasting the health care provider's time.

I visited the GP for a diagnosis when the thought occurred to me: there is really something physically wrong with me, this is not all in my head or the consequence of being alone with two kids making everything difficult, difficult, difficult. (Female, 49 years)

I remember that I felt guilty for taking his time, and I felt uncomfortable in the waiting room among the 'real patients'. (Female, 47 years)

The patient's attitude towards health care appeared to be of importance. Patients think differently about consulting health care providers. Previous negative experiences in the communication with health care providers might influence the decision to return to the doctor with symptoms.

I grew up in an environment that favors alternative medicine, and thus did not seek help from mainstream medicine until later on. (Female, 70 years)

[…] Pain in my right leg and the lower back. Physical therapy. […] Still tired. Sometimes a severe cramping around my heart. […] In June […] extensive testing, diagnosis of angina pectoris. Walking becomes more difficult: dizziness, my handwriting becomes spidery. Now and then blood tests, the results are always normal. My GP: "There is nothing wrong with you." But where does this fatigue come from then? GP: "Well, I'm not sure. But you're not that young anymore!" I felt really tired lately, especially in the morning when getting out of bed. I get depressed. (Female, 84 years)

Some patients described they adapted their lifestyle or made adjustments in order to relieve the symptoms.

My left hand started to shake in more and more situations, it was especially bad in the choir; new sheet music in my hands, reading music and lyrics, and singing at the same time. […] At the December concert, I sang with a home-made brace to control my hand. (Female, 53 years)

Patients indicated that they altered their decision to seek help when the symptoms got worse, new symptoms appeared or symptoms did not recover spontaneously. The restrictive influence of symptoms on the patient's daily life was also mentioned as a reason to visit a health care provider.

After a good holiday with my family, the tremor still continued, so I visited the GP again. (Male, 32 years)

At one point, I became tired very easily, I lost strength in my arms. I was a production worker and noticed I had more and more trouble with my motor skills, then I went to the GP. (Male, 42 years)

A few patients actively decided not to seek help yet, because of fear for the diagnosis.

The first symptom was a tremor of the right hand, which came about suddenly. Furthermore, my husband and friends noticed that I did everything slower. For me, the worst part was the feelings of depression; that was not my nature. I had everything I ever wanted, but I could not enjoy it, it was horrible. My handwriting had changed as well, it became very small.

I struggled with it for two years, but deep in my heart I knew it was not good and that I probably had Parkinson's disease. (Female, 61 years)

The process of diagnosing PD

Some patients described the GP immediately noticed that their symptoms could be signs of PD and referred them to the neurologist, who instantly diagnosed the disease. However, more than half of the patients mentioned their GP did not recognize the symptoms, sometimes not even when the patient specifically asked if it could signal PD. Other patients described they were referred to the physical therapist or orthopedic surgeon without a clarified diagnosis. Some patients mention their neurologist seemed to have difficulties recognizing the symptoms.

My fingers no longer cooperated. I could not hold a pen and had constipation. Then I visited the GP. The GP did not recognize it as PD. He said the problems were caused by aging. (Male, 69 years)

I had shoulder complaints. I went to the GP, who referred me to the orthopedic surgeon. He told me I had bursitis, which was treated by injections in my shoulder. I had to come back every 6 weeks […]. I was treated for 2 years like this. (Male, 52 years)

Reflecting on the pathway

Patients reflected differently on their diagnostic pathway. Nearly half of the patients believed they could not have been diagnosed earlier.

I do not think I could have been diagnosed faster. My complaints are always taken seriously, even when they were 'vague'. Some can, retrospectively, be attributed to PD. (Male, 83 years)

However, the majority of the patients did believe their diagnosis was delayed. A few patients felt this was the result of their own help-seeking behavior. Others described that they believed the health care provider did not recognize their symptoms in time or postponed referral to a neurologist. Nevertheless, most patients were satisfied with the trajectory towards diagnosis. A few discontent patients described difficulties in the communication with their doctor.

If I look back, then the diagnosis probably could have been made earlier, but this was my doing. But I do not regret the path I took and that I only took action after 2 years. (Female, 61 years)

If I had gone to the GP earlier, and the GP had known more about PD, then some things could have been detected faster. If the diagnosis was made earlier, I could have felt better about it emotionally. (Male, 53 years)

Discussion and conclusion

Discussion

To the best of our knowledge, this is the first study that explores the pathway to the diagnosis of PD from the patients' viewpoint. The pathway to the diagnosis of PD, as described by patients, can be divided into three time intervals: recognition of the symptoms, the decision to seek help and the process of diagnosing PD. Impeding and stimulating factors concerning the patient, the health care provider and the disease itself can influence each of these time intervals. Although more than half of the patients believed their diagnosis was delayed, the majority of all patients were satisfied with the trajectory towards diagnosis.

Earlier studies have shown that the pathway to the diagnosis of a disease can be divided into several stages in which delay can appear. Safer et al. proposed a model of three stages of delay in seeking care at a medical clinic.[19] Andersen et al. and Walter et al. have built further on this, resulting in the model of Walter that contains four intervals with clearly defined start and end-points: the appraisal interval (from detection of bodily change(s) to perceiving a reason to discuss symptoms with a GP); the help-seeking interval (from perceiving a reason to visit the GP to the first consultation with a GP); the diagnostic interval (from the first appointment to the diagnosis); and the pre-treatment interval (the time between the diagnosis and initiation of treatment).[20, 21] With the exception of the pre-treatment interval, this model seems very well applicable to our results.

According to the model of Walter et al.[21], the intervals of the pathway to diagnosis are influenced by patient -, health care provider- and disease-related factors. Most influencing factors found in our study are patient-related and can be stimulating as well as impeding. These include (lack of) knowledge of PD, alternative explanations for the symptoms, assessment of the severity of the symptoms (and possible interpretation as common illnesses), the influence of family, friends and colleagues, adaptations in lifestyle to relieve the hinder and the restrictive influence on daily life. These results are in line with the factors found to be of influence in studies on cancer and diabetes.[16, 17, 22] However, our study reveals that the media play an important role as well on the diagnostic pathway of PD. Books and television paint a classic picture of PD, thereby limiting the knowledge of the disease and influencing the assessment of severity. Furthermore, our findings reveal that fear for the diagnosis can hold-back patients from seeking help. Although this fear is described in cancer research as well, fear for cancer might be difficult to compare to fear for PD. Patients associate cancer with painful treatments and death[22] while they might not have such explicit ideas about PD. Since most prodromal symptoms of PD are not acute or life threatening, patients can decide to postpone seeking help. Our study showed that the patient's attitude towards health care providers, sometimes prompted by earlier experiences, can also influence the decision to seek help. Some patients mention they are hesitant to present their non-specific symptoms to their GP, afraid they might be seen as somatizers. This is in line with earlier research that showed that patients carefully consider when to consult their GP and are concerned about

going with non-specific symptoms.[16, 17, 22, 23] Research in epilepsy also showed that patients might postpone seeking help because they are not ready to accept the diagnosis of a chronic disease.[24] The same might be true for patients with PD.

A number of health care provider-related factors influence the diagnostic pathway as well. Health care providers need to recognize the (prodromal) symptoms and suspect PD. The importance of educating physicians to consider the possibility of prodromal PD is already expressed in earlier research.[25] However communication also seems to be an important factor influencing patients' contentment with their diagnostic pathway.[26] It is desirable that patients are provided with customized information concerning the suspected diagnosis and treatment options. They should be encouraged to participate in the decision-making process on referral to a movement disorder specialist and on therapy.[14, 27] For this, physicians need to persuade patients to ask questions, articulate their expectations and voice their preferences.[28]

Finally, disease-related factors are of influence on the diagnostic pathway, in particular on the decision to seek help. In line with literature[16, 17, 22], we found that patients are more inclined to seek help when their symptoms become worse or do not recover spontaneously. However, this requires patients who are aware of bodily changes and are capable to carefully monitor their symptoms.[19] In addition, the general population should be more aware of symptoms that can be highly relevant for the early detection of PD.[25]

The pathway to diagnosis is a dynamic and iterative process in which patients may not experience a linear passage through the intervals, and in which steps can be repeated until PD is diagnosed. This is also shown in the model of Walter et al.[21] Based on that model we developed a figure that summarizes all the influencing factors found in this study, as discussed above (Figure 2).

Asking patients to retrospectively describe their diagnostic pathway inevitably leads to limitations. Patients may find it difficult to remember precisely what took place prior to the diagnosis, at what time and in which order, especially since for some patients it is years ago since they were diagnosed. Therefore, recall bias cannot be ruled out. However, with the use of a digital essay format we gave patients the opportunity to go back in time and recall the pathway, while supporting the arrangement of their memories. The format with open questions led to essays of comparable structure, at the same time maintaining the possibility for the patients to individualize their answers. Unfortunately, the time frame in the essays is not always clear. Therefore there are limitations in comparing the time frame of each interval with other studies.[9, 26]

The preset questions and the approach via internet have other disadvantages. The extent of details found in the essays is limited; in-depth interviews could have given more detailed

information on the diagnostic pathway. Moreover, it is estimated that less than 25% of all patients with PD in the Netherlands is a member of the Dutch Parkinson's Disease Association. Furthermore, only a selection of the patient members of the Association is reached with the approach through email and the use of a digital essay format. Members who are unable to use a computer or feel uncomfortable with it are left out. Although information on the demographic variables of the non-responders and the reasons why they did not take part is lacking, the inability or undesirability to use the computer may be the explanation that the respondents, whose essays are included in the analysis, are relatively young and well educated despite the application of purposive sampling. It cannot be ruled out that the results of our study are influenced by this, since it is known that health literacy is influenced by the patient's level of education and lifestyle commitments.[29, 30] However, we included a highly variable sample of respondents and achieved saturation in the analysis. Moreover we verified the figure and the position of the themes within it with the analysis of a second sample of essays. Therefore we feel confident that our results hold sufficient external validity.

Conclusion

The pathway to the diagnosis of PD is a dynamic and an iterative process. As described by patients, both patients and GPs have difficulties in recognizing the early symptoms of PD.
Patients often have a limited perception of PD, based on public figures with an advanced stage of PD. More than half of the patients believed they could have been diagnosed earlier. At the same time, the majority of all patients are content with their pathway. Nevertheless, patients can benefit a lot from an early diagnosis and timely therapeutic intervention, taking into account the patient's personal situation and preferences. Therefore it is important that patients, their significant others and GPs learn to recognize the early symptoms of PD and act accordingly.

Practice implications

In order to facilitate an earlier diagnosis of PD, which enables shared decision-making between patients and health care providers, educating the general population on possible prodromal symptoms of PD should be considered. The image of PD, as it is spread by the media, has to be modified from the classic image of the old man with advanced symptoms to a more complete representation of the disease.

It is necessary to explore whether the patient's view on possible lack of knowledge of the GP is underlined by the health care providers themselves. The GP has a central role in putting together all pieces of the puzzle which a patient has presented over time, in order to signal PD. Furthermore, he/she is the gatekeeper to care, who has to refer to a neurologist. Therefore, it seems without debate that knowledge on the prodromal symptoms of PD is essential. A subsequent quantitative analysis of all 900 collected essays of patients with PD is necessary and will give us more insight into the prodromal symptoms of PD that are experienced by patients and reported spontaneously by them.

Finally, we would strongly advise to no longer use 'stage(s) of delay' in the conversation concerning a diagnostic process. Delay has a negative connotation and does not appreciate the autonomy of the patient, who might decide to hold-back help-seeking for a number of reasons. Furthermore, our results show that most patients are content with the pathway they experienced, although objectively spoken delay might have taken place. In other literature the term 'time interval' is suggested as an alternative for 'stage of delay'.[21] We believe this term accurately describes a stage (the time between two events) and, most importantly, does not criticize the decisions made by patients, alone or together with their health care providers.

Ethics committee approval and informed consent

The research ethics committee of the Radboud university medical center studied the protocol of the study and concluded that the study can be carried out in the Netherlands without an approval by the regional accredited research ethics committee (11-12-2013).

The authors confirm that all personal identifiers have been removed or disguised so the patient(s) described are not identifiable and cannot be identified through the details of the story.

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3

The diagnostic pathway of Parkinson's disease: a cross-sectional survey study of factors influencing patient dissatisfaction

I could have been diagnosed earlier if my GP had made the referral earlier. The advice of our GP to put Parkinson's out of our heads was especially upsetting; my husband and I [...] were extremely overwhelmed by the diagnosis of the neurologist. Because of this, we felt anger towards the GP [...], and our trust in her has been damaged. (Female, 58 years)

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Submitted

Abstract

Background: The diagnostic pathway of Parkinson's disease (PD) is often complicated. Experiences during this pathway can affect patients' satisfaction and their confidence and trust in health care providers. Although health care providers cannot influence the impact of the diagnosis, they can influence how patients experience the pathway. This study, therefore, aims to provide insight into PD patients' dissatisfaction with the diagnostic pathway and to describe the factors that influence it.

Methods: We carried out a cross-sectional survey study among 902 patient members of the Dutch Parkinson's Disease Association, who were each asked to write an essay about their diagnostic pathway. A coding format was developed to examine the content of these essays. Inter-observer agreement on coding patient dissatisfaction was calculated using Cohen's kappa. The chi-square test and a multivariate logistic regression analysis were performed to assess the relation between dissatisfaction and sex, level of education, duration of the pathway, communication with the general practitioner (GP) and the neurologist, the number of health care providers involved, whether or not a second opinion had taken place (including the person who initiated it) and diagnostic delay (taking into consideration who caused the delay according to the patient). A subgroup analysis was performed to gain insight into sex-related differences.

Results: Of all patients, 16.4% explicitly described they were dissatisfied with the diagnostic pathway, whereas 4.8% were very satisfied. The inter-observer agreement on coding dissatisfaction was $\kappa = 0.82$. The chance of dissatisfaction increased with a lower level of education, the involvement of more than one additional health care provider, a second opinion initiated by the patient and delay caused by a health care provider. When only the GP and the neurologist were involved, women were more likely to be dissatisfied than men.

Conclusions: PD patients' dissatisfaction with the diagnostic pathway is related to a lower level of education, a second opinion initiated by the patient and experienced diagnostic delay. GPs can positively influence patients' experiences if they are aware of these risk factors for dissatisfaction and pay extra attention to communication and shared decision-making. This will contribute to a trusting therapeutic relationship that is indispensable with progression of the disease.

Background

Patients' experiences during the pathway to a diagnosis can influence long-term care. Research in cancer patients shows that patients tend to lose confidence and trust in their general practitioner (GP) if the nature of the presented symptoms is not immediately recognized by the GP and if multiple visits to the GP are necessary before referral takes place.[1-3] Moreover, patients may interpret the need to consult their GP repeatedly as lack of responsiveness and thus as delay caused by the GP, resulting in patient dissatisfaction.[4]

Diagnosis timing issues may induce patients to change general practice.[5] The importance of timeliness of diagnosis related to patient dissatisfaction emerges from a study amongst cancer patients, showing that patients suffering from cancer types that are difficult to recognize, such as ovarian cancer and multiple myeloma, are more likely to change practice than patients with types of cancer that are easier to recognize, such as melanoma and breast cancer.[5] In addition, long-term care experiences of cancer patients are worse for those who visited their GP several times before they were referred than for those who were referred instantly.[2]

Parkinson's disease (PD) is a progressive neurodegenerative disorder that can be difficult to diagnose.[6] Classic symptoms such as muscular rigidity and tremor are not always present and may be preceded by a variety of motor and non-motor symptoms that are not necessarily disease-specific.[7-10] When patients consult their GP, symptoms are often still limited and embedded in clinical uncertainty, while referral to the neurologist takes place later in the disease trajectory.[10-12] This may explain the difficulties GPs encounter in recognizing PD as the common cause of these symptoms and in referring accordingly.[10, 12] As a consequence, the pathway to the diagnosis of PD can be lengthy and uncertain and, unless well explained, it is reasonable to expect a negative influence on patient confidence, trust and satisfaction.[6, 13]

Although the impact of a PD diagnosis cannot be taken away completely, health care providers can have an influence on how patients experience the diagnostic pathway. It is known that PD patients' dissatisfaction with the way the diagnosis of PD is explained to them has an impact on health-related quality of life.[14] Lack of involvement in therapy decisions is also negatively related to satisfaction and compliance with therapy.[15] However, research into patient experiences of the diagnostic pathway of PD is limited and does not provide any insight into factors influencing patient dissatisfaction.[13, 16] Patients will benefit from a sustained trusting relationship with their GPs, in which they have confidence in the personal care provided by the GP, as progression of the disease will inevitably cause health problems that require the GP's involvement.[17] In order to optimize patients' experiences of the pathway to the diagnosis of PD and, hence, to contribute to a trusting patient-doctor relationship, this study aims to improve our understanding of PD patients' dissatisfaction with the diagnostic pathway and to describe the factors influencing it.

Methods

Recruitment and data collection

We conducted a cross-sectional survey study among patient members of the Dutch Parkinson's Disease Association. All members with a known email address (n = 4717) were approached digitally to enlist their participation. Patients were asked to fill in their demographic characteristics at the time of diagnosis: sex, age, highest level of education finished, employment status and civil status. They were also asked to describe their experiences of the pathway from the first recognizable symptom(s) to the diagnosis of PD. To facilitate patients in formulating their essay, we provided them with a number of guiding questions that were based on the literature and expert opinion and had been tested in a pilot study (Table 1).

	Question
1.	Can you describe the first symptom(s) that eventually turned out to be a forerunner sign of PD? What did you do when you experienced this symptom or these symptoms?
2.	Can you describe what happened next, until the moment you were diagnosed with PD?
3.	What role was there for people in your surroundings during the diagnostic pathway?
4.	Looking back on the diagnostic pathway, how do you feel about the timing of the diagnosis? Can you describe the consequences of this timing for you and your family?

Table 1. Questions guiding patients to describe their experiences of the diagnostic pathway of Parkinson's disease

In case patients had questions, concerns or hesitations, they could contact the researcher (AP). After finishing their essays, patients had to agree to submission, a step that was assessed as informed consent. Participation was one-time only, voluntarily and anonymous. The research ethics committee of the Radboud university medical center examined the protocol of the study and concluded that the study could be carried out in the Netherlands without needing approval by the regional research ethics committee.

A qualitative analysis of a purposive sample of 52 essays preceded this study. We refer to the paper describing this analysis for more detailed information on recruitment, data collection and results.[16] The qualitative analysis results were used to create a format to examine the content of all essays. Details on the coding format are described in Additional file 1.

Though patient dissatisfaction with the diagnostic pathway was the main focus of the current study, patients were not explicitly asked for their satisfaction or dissatisfaction. Rather, we encouraged them to describe their feelings about the timing of the diagnosis and the consequences of this timing in order to gain insight into patients' spontaneous reporting of the diagnostic pathway (Table 1). We only applied codes if patients spontaneously and unmistakably expressed their satisfaction or dissatisfaction: 'satisfied' was coded when

patients were clearly positive, and 'dissatisfied' was coded when patients explicitly mentioned problems or made negative remarks. All other cases were coded 'neutral'. To enable the researchers to interpret satisfaction and dissatisfaction with the diagnostic pathway, it was defined as 'the overall feeling a patient expressed about the diagnostic pathway in his/her essay', and it was independently coded by two researchers (AP, OdB) in a random sample of 225 essays (25%) to enable calculation of inter-observer agreement.

The same researchers also independently coded 154 essays (17.1%) completely, initially to create consensus on the coding method, and later to discuss doubts in coding. The other 748 essays were coded by one researcher (OdB). Codes were only applied if patients in their essay explicitly described the duration of the pathway, communication with the GP or the neurologist, the number of different health care providers involved, a second opinion or experienced delay.

Data analysis

Statistical analyses were conducted using SPSS 22.0. Descriptive statistics were calculated. As patient dissatisfaction was the main focus of our analysis, the expressed feelings were divided into two categories: dissatisfied and neutral/satisfied. Cohen's kappa was used to calculate inter-observer agreement.

The chi-square test was used to assess the relation between dissatisfaction with the diagnostic pathway and factors that may have been of influence. These factors included the demographic variables sex and level of education, the latter divided into low (primary school/ vocational education), medium (secondary school) and high (higher professional education/ university). Moreover, we assessed the relation between dissatisfaction and duration of the diagnostic pathway (divided into unknown, <2 years or ≥2 years on the basis of the literature[6, 8, 9]); communication with the GP or the neurologist (negative, neutral/positive); and the number of different health care providers involved (0, 1, 2, ≥3). As guidelines in the Netherlands describe the involvement of a GP and a neurologist as usual care in the pathway to the diagnosis of PD, these health care providers were excluded from the number of health care providers involved.[11, 18]

In addition, we performed the chi-square test to assess the relation between dissatisfaction and second opinion. Second opinion was defined as 'the involvement of a second neurologist during the pathway towards the diagnosis of PD' and was categorized into: no second opinion/ not mentioned; second opinion on the patient's initiative (including the combined initiative of patient and health care provider); and second opinion on the health care provider's initiative. We also assessed the relation between dissatisfaction and experienced diagnostic delay, taking into consideration who caused the delay according to the patient. Delay was divided into: no delay; not (clearly) mentioned; caused by the patient (including caused by both the patient and the health care provider); caused by the health care provider(s); and unknown who caused it.

A multivariate logistic regression analysis was performed to assess the independent association between dissatisfaction and sex, level of education, duration of the diagnostic pathway, the number of different health care providers involved, second opinion and experienced delay. As only few patients explicitly described their communication with the GP and the neurologist, this factor was excluded from the regression analysis. We also excluded second opinions if it was unknown on whose initiative they had taken place. As the literature shows that female patients tend to be more dissatisfied with care than male patients[19], we also performed a subgroup analysis to gain insight into possible sex-related differences. Therefore, we added interaction terms of sex with the other variables to the multivariate regression model. A P-value less than 0.05 was considered statistically significant.

Results and discussion

Characteristics of the study population

Of all patient members who received an email, 27% started and 21% finished the essay. Seventy-two essays were excluded due to incorrect or uncertain diagnosis of PD or a complete lack of information. Finally, 902 essays were included in this study (Figure 1). More men than women participated, and most patients had a high level of education (Table 2). Mean age at the time of diagnosis was 60 years (SD 9.9).

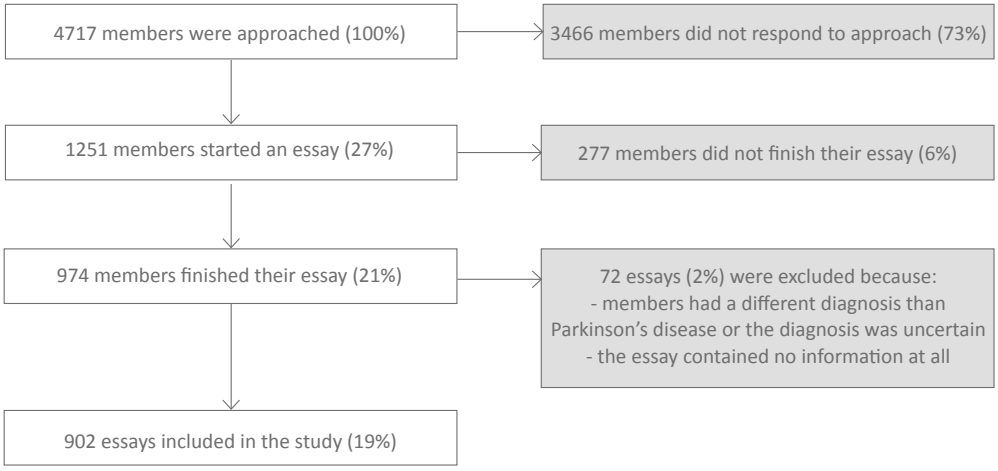

Figure 1. From recruitment of patient members of the Dutch Parkinson's Disease Association to essays included

Demographic variable		n = 902 (100%)
Sex	Male	550 (61.0%)
	Female	352 (39.0%)
Mean age in years (SD)	60.1 (9.9)	
Level of education	Low	250 (27.7%)
	Medium	284 (31.5%)
	High	368 (40.8%)
Employment	Employed	352 (39.0%)
	Self-employed	71 (7.9%)
	Retired	307 (34.0%)
	Recipient of sickness benefits	18 (2.0%)
	Unemployed	86 (9.5%)
	Combination of employments/other	68 (7.5%)
Civil status	Single*	80 (8.9%)
	With partner	596 (66.1%)
	With family (including partner)	217 (24.1%)
	Other	9 (1.0%)

*Including widowed and divorced

Table 2 Included PD patients' characteristics at the time of diagnosis

Patient dissatisfaction with the diagnostic pathway

The inter-observer agreement of the two researchers (AP, OdB) on coding patient dissatisfaction with the diagnostic pathway was $\kappa = 0.82$ (95% CI 0.72-0.91).

More than one in seven patients (n = 148; 16.4%) explicitly described they were dissatisfied with the experienced diagnostic pathway. Most patients (n = 711; 78.8%) did not clearly express their opinion, while less than five per cent (n = 43; 4.8%) mentioned they were satisfied.

Dissatisfaction was significantly associated with several factors. Female sex ($P < 0.01$), duration of the pathway ($P < 0.001$), communication with the GP or the neurologist ($P < 0.001$ and $P < 0.01$, respectively), the number of health care providers involved ($P < 0.001$), second opinion ($P < 0.001$) and experienced delay ($P < 0.001$) increased the chance of dissatisfaction (Table 3).

Variable	Patient dissatisfaction n (%)	Patient satisfaction/neutral n (%)	P-value
Sex (n = 902)			<0.01*
- Male	74 (13.5%)	476 (86.5%)	
- Female	74 (21.0%)	278 (79.9%)	
Level of education (n = 902)			0.22
- Low	49 (19.6%)	201 (80.4%)	
- Medium	40 (14.1%)	244 (85.9%)	
- High	59 (16.0%)	309 (84.0%)	
Duration of the diagnostic pathway (n = 902)			<0.001*
- Unknown	54 (13.3%)	352 (86.7%)	
- <2 years	28 (12.1%)	204 (87.9%)	
- ≥ 2 years	66 (25.0%)	198 (75.0%)	
Communication with the general practitioner (n = 77)			<0.001*
- Negative	40 (69.0%)	18 (31.0%)	
- Neutral/positive	2 (10.5%)	17 (89.5%)	
Communication with the neurologist (n = 78)			<0.01*
- Negative	41 (62.1%)	25 (37.9%)	
- Neutral/positive	1 (8.3%)	11 (91.7%)	
Number of health care providers involved (n = 902)[a]			<0.001*
- 0	43 (8.2%)	484 (91.8%)	
- 1	52 (21.3%)	192 (78.7%)	
- 2	31 (34.8%)	58 (65.2%)	
- ≥3	22 (52.4%)	20 (47.6%)	
Second opinion (n = 856)[b]			<0.001*
- No/not mentioned	93 (12.5%)	650 (87.5%)	
- Yes, on the patient's initiative	29 (45.3%)	35 (54.7%)	
- Yes, on the health care provider's initiative	12 (24.5%)	37 (75.5%)	
Experienced delay (n = 902)			<0.001*
- No delay	15 (3.2%)	454 (96.8%)	
- Not (clearly) mentioned	9 (6.8%)	124 (93.2%)	
- Yes, caused by the patient	2 (2.6%)	74 (97.4%)	
- Yes, caused by a health care provider	111 (61.0%)	71 (39.0%)	
- Yes, unknown who caused it	11 (26.2%)	31 (73.8%)	

*Statistically significant, P < 0.05
[a]Excluding GP and neurologist
[b]Excluding second opinion, initiative unknown

Table 3. Univariate analysis of factors influencing patient dissatisfaction with the diagnostic pathway of Parkinson's disease

The multivariate analysis showed that low-educated patients were more likely to be dissatisfied than medium and high-educated patients (OR 0.45; CI 0.2-0.9 and OR 0.46; CI 0.2-0.9, respectively). The chance of dissatisfaction was also significantly higher when more than one additional health care provider was involved. With the involvement of two extra health care providers, the odds ratio for patient dissatisfaction was 2.53 (CI 1.2-5.3) compared to a situation in which only the GP and the neurologist were involved. If three or more additional health care providers were involved, the odds ratio for dissatisfaction was even higher (OR 3.92; CI 1.4-10.7). A second opinion on the patient's initiative increased the chance of dissatisfaction compared to cases without a second opinion (OR 5.04; CI 2.3-10.9). In addition, when patients experienced delay caused by a health care provider, they were significantly more likely to be dissatisfied than patients who did not experience delay (OR 38.78; CI 20.0-75.0) (Table 4).

Variable (n = 856)[b]	Odds ratio (OR) for dissatisfaction	95% Confidence Interval (CI)	P-value
Sex			0.12
- Male	Reference		
- Female	1.50	0.9-2.5	0.12
Level of education			0.02*
- Low	Reference		
- Medium	0.45	0.2-0.9	0.02*
- High	0.46	0.2-0.9	0.01*
Duration of the diagnostic pathway			0.47
- Unknown	Reference		
- <2 years	1.20	0.6-2.4	0.61
- ≥ 2 years	1.43	0.8-2.5	0.22
Number of health care providers involved[a]			0.01*
- 0	Reference		
- 1	1.66	0.9-3.0	0.09
- 2	2.53	1.2-5.3	0.01*
- ≥3	3.92	1.4-10.7	<0.01*
Second opinion			<0.001*
- No/not mentioned	Reference		
- Yes, on the patient's initiative	5.04	2.3-10.9	<0.001*
- Yes, on the health care provider's initiative	2.11	0.8-5.4	0.12
Experienced delay			<0.001*
- No delay	Reference		
- Not (clearly) mentioned	1.85	0.7-4.6	0.19
- Yes, caused by the patient	0.84	0.2-4.0	0.83
- Yes, caused by a health care provider	38.78	20.0-75.0	<0.001*
- Yes, unknown who caused it	7.14	2.7-19.0	<0.001*

*Statistically significant, P < 0.05
[a]Excluding GP and neurologist
[b]Excluding second opinion, initiative unknown

Table 4. Multivariate logistic regression of factors influencing patient dissatisfaction with the diagnostic pathway of Parkinson's disease

Men and women significantly differed in the relation between dissatisfaction with the diagnostic pathway and the number of health care providers involved during this pathway. If no health care providers were involved, female patients were more likely to be dissatisfied than male patients (OR 3.11; CI 1.4-7.0). There were no significant differences between men and women in the chance of dissatisfaction with the involvement of one or more additional health care provider(s) (OR 0.58; CI 0.2-1.4 with one, OR 1.63; CI 0.4-5.8 with two, and OR 1.92; CI 0.3-12.1 with three or more health care providers involved). No interaction effects were found for any other variables (results provided in Additional file 2).

Discussion

Most patients in our study do not describe dissatisfaction with the diagnostic pathway of PD. However, more than one in seven patients is explicitly dissatisfied. The chance of dissatisfaction is increased with a lower level of education, the involvement of more than one additional health care provider, a second opinion on the patient's initiative and delay caused by the health care provider. In addition, if only the GP and neurologist have been involved in the diagnostic pathway, women are more likely to be dissatisfied than men.

Although dissatisfaction with the diagnostic pathway of PD appears to be playing a limited role in quantitative terms, dissatisfaction with the initial diagnostic process can have an impact on long-term care, stressing the importance of paying attention to it: cognitions formed at an early stage tend to determine care experiences during the further treatment episode.[2, 5, 20]

Our study shows that it is not the duration of the diagnostic pathway of PD on its own that leads to patient dissatisfaction, but that other factors appear to be important as well. In line with the literature, the women in our study are more likely to be dissatisfied than the men.[5, 19] Contrary to what we expected, however, the chance of dissatisfaction is highest for low-educated patients.[5, 19] As only few patients in our study explicitly mention their communicative experiences with the GP and the neurologist, we could not demonstrate an independent association between dissatisfaction and communication. It is likely, though, that communication at least partly explains the finding that low-educated patients are more often dissatisfied than patients with higher education as low-educated PD patients are known to have a lower level of health literacy than high-educated patients and a low level of health literacy negatively influences patients' ability to obtain and understand information about a disease.[21, 22]

In the case of PD, the complexity and abstractness of the process in the brain that causes the complaints and the varying expression of the disease can be difficult for a doctor to explain understandably and challenging for a patient to grasp fully. An earlier study shows that the way the diagnosis of PD is communicated to a patient is very important.[13] Moreover, the difficulty for GPs to recognize PD and the fact that a diagnosis is not 100% certain until autopsy is performed are likely to influence patients' experiences of the diagnostic pathway of PD as well.[10, 17] Cancer patients, for example, mention they feel uncertain and anxious if referral is not explained carefully, and patients with amyotrophic lateral sclerosis report falsely raised hopes after every negative investigation.[20, 23] Our study shows that patients who feel that their health care provider is responsible for delay in the diagnostic pathway are more likely to be dissatisfied than patients who do not describe delay.

Lack of communication during the diagnostic pathway is also known to have a long-term impact on the patient-doctor relationship, a relationship that depends on patients' satisfaction and their confidence and trust in the health care provider - with the risk of disappointment as the distinguishing feature between the latter two: in a situation of trust one is more likely to be disappointed-.[20, 24] In our study, patient dissatisfaction is related to a second opinion on the patient's initiative. Although we do not know whether dissatisfaction is the cause or the result of the patient's request for a second opinion, earlier research shows that dissatisfaction is negatively associated with trust and that trust limits the tendency for patients to request for a second opinion.[24] Physicians who listen carefully, behave empathically and communicate clearly are more likely to be trusted by their patients.[24, 25] Moreover, trust is further enhanced if patients feel they are treated as equal partners.[24, 25]

Though the negative impact of being diagnosed with PD cannot be ruled out, GPs can contribute to their patients' experiences of the diagnostic pathway of PD in a positive way by using their central role in symptom recognition and referral to communicate openly about the clinical uncertainty involved in PD diagnosis and about the expectations of referral, while taking into account a patient's level of health literacy and offering scope for questions, hesitations and emotions. This is how GPs and patients can go through the diagnostic pathway of PD together and make shared decisions whenever possible or desirable. It is likely that such an experience will contribute to patient satisfaction with the pathway and will help to maintain a trusting therapeutic relationship that is indispensable with progression of the disease.[17]

Strengths and limitations of the study

To the best of our knowledge, this is the first study that reports on patient dissatisfaction with the diagnostic pathway of PD and factors that might be of influence. We included a large number of essays. Moreover, we used an original approach to mixed methods research.[26] Our data are based on patients' spontaneous reporting, thus reflecting what matters most to them, rather than reflecting their recognition of pre-determined items.

We used a coding format that was based on the results of our preceding qualitative study, and a considerable part of all essays was independently coded by two researchers, who reached consensus in case of coding disagreement.[16] The strength of the inter-observer agreement on coding dissatisfaction can be considered 'almost perfect', confirming our opinion that we used a reliable method to extract the content of the essays.[27] We feel confident that the results of our study provide valuable new information that can be used to improve patient experiences of the diagnostic pathway of PD.

Nevertheless, there are limitations to consider when interpreting the results of this study. As patients described their experiences retrospectively, recall bias cannot be ruled out. In addition, this study used the diagnostic pathway of PD as a starting-point, yet patients are likely to have had previous care experiences with their GPs, and their satisfaction or dissatisfaction with these earlier care episodes may have influenced their experiences with the diagnostic pathway of PD as well as their description of it. In addition, dissatisfaction with the pathway cannot solely be interpreted as dissatisfaction with the GP, as more health care providers have likely been involved in the diagnostic process. The spontaneous reporting method does not allow for interpretation of causality and if information on a second opinion is lacking, for example, it may not have been performed or it may not have been reported.

For practical reasons, we chose to approach only those PD patients in the Netherlands who are members of the Dutch Parkinson's Disease Association, and not all patients we approached finalized their essay. As information on the non-responders and the patients who did not finish their essay is lacking, selection bias cannot be ruled out. As a possible result of our decision to use a digital approach, the patients included in our study are relatively young,

and it cannot be ruled out that this unequal distribution has influenced the results as well, as younger patients are generally known to be less satisfied.[19]

Conclusions

The diagnostic pathway of PD can be lengthy and uncertain, and more than one in seven PD patients is clearly dissatisfied with it. This study shows that patient dissatisfaction is related to a lower level of education, a second opinion on the patient's initiative and delay that is caused by the health care provider according to the patient. GPs can positively influence their patients' experiences if they are more aware of these risk factors for dissatisfaction and pay extra attention to open communication on the clinical uncertainties of the early symptoms of PD and on shared decision-making on referral. This is likely to contribute to a trusting relationship between PD patients and their GPs, a relationship that is essential at all stages of the disease.

Ethics

The research ethics committee of the Radboud university medical center examined the protocol of the study and concluded that it could be carried out in the Netherlands without requiring approval by the regional research ethics committee (December 11th 2013).
Patient participation was voluntarily and anonymous and patients' explicit permission to submit the essay was assessed as informed consent with participation.

Acknowledgements

The authors thank the patient members of the Dutch Parkinson's Disease Association for their participation.

Additional files

Additional file 1

Coding format to examine patients' essay contents; only variables included in the analysis have been incorporated

Variable	Values
Sex	1 = Male 2 = Female
Year of birth	Year
Year of diagnosis	Year
Age at the time of diagnosis	Age in years
Highest level of education finished	Low = Primary school/Vocational education Medium = Secondary school High = Higher professional education/university
Employment status at the time of diagnosis	1 = Employed 2 = Self-employed 3 = Retired 4 = Recipient of sickness benefits 5 = Unemployed 99 = Combination of employments/other
Civil status at the time of diagnosis	1 = Single (including widowed and divorced) 2 = With partner 3 = With family (including partner) 99 = Other
Duration of the diagnostic pathway	0 = unknown 1 < 2 years 2 ≥ 2 years
Communication with the GP during the diagnostic pathway	0 = Not mentioned/unknown 1 = Negative 2 = Neutral/Positive
Communication with the neurologist during the diagnostic pathway	0 = Not mentioned/unknown 1 = Negative 2 = Neutral/Positive
Health care providers involved in the diagnostic pathway	Each health care provider as a separate variable 0 = Not involved/not mentioned 1 = Involved
Second opinion	0 = No/not mentioned 1 = Yes, on the initiative of the patient or patient and health care provider(s) 2 = Yes on the initiative of health care provider(s) 99 = Yes, initiative unknown
Experienced delay during the diagnostic pathway	0 = No 1 = Not (clearly) mentioned 2 = Yes, caused by the patient or by both patient and health care provider(s) 3 = Yes, caused by health care provider(s) 4 = Yes, unknown who caused it
Satisfaction with the diagnostic pathway	1 = Explicitly dissatisfied 2 = Neutral 3 = Explicitly satisfied

Additional file 2

Multivariate logistic regression of factors influencing patient dissatisfaction with the diagnostic pathway of Parkinson's disease, including interaction terms of sex with other variables

Variable (n = 856)[b]	Odds ratio (OR) for dissatisfaction	95% Confidence Interval (CI)	P-value
Sex			0.20
- Male	Reference		
- Female	2.63	0.6-11.6	0.20
Level of education			0.07
- Low	Reference		
- Medium	0.41	0.2-1.1	0.08
- High	0.34	0.1-0.9	0.02*
Duration of the diagnostic pathway			0.94
- Unknown	Reference		
- <2 years	1.17	0.4-3.2	0.76
- ≥ 2 years	1.13	0.5-2.5	0.77
Number of health care providers involved[a]			<0.01*
- 0	Reference		
- 1	3.86	1.7-9.0	<0.01*
- 2	3.59	1.3-9.8	0.01*
- ≥3	5.40	1.3-21.9	0.02*
Second opinion			<0.01*
- No/not mentioned	Reference		
- Yes, on the patient's initiative	7.44	2.3-24.5	<0.01*
- Yes, on the health care provider's initiative	3.72	1.0-13.8	<0.05*
Experienced delay			<0.001*
- No delay	Reference		
- Not (clearly) mentioned	2.15	0.6-8.2	0.26
- Yes, caused by the patient	1.49	0.2-13.2	0.72
- Yes, caused by a health care provider	50.78	19.4-133.0	<0.001*
- Yes, unknown who caused it	1.74	0.2-16.4	0.63
Level of Education x Sex			0.59
- Low x Male	Reference		
- Medium x Male	0.95	0.2-3.8	0.94
- High x Male	1.74	0.5-6.4	0.40
Duration of the diagnostic pathway x Sex			0.51
- Unknown x Male	Reference		
- <2 years x Male	1.13	0.3-4.8	0.87
- ≥ 2 years x Male	1.99	0.6-6.6	0.26
Number of health care providers involved[a] x Sex			**0.04***
- 0 x Male	Reference		
- 1 x Male	0.16	0.0-0.6	<0.01*
- 2 x Male	0.40	0.1-2.0	0.26
- ≥3 x Male	0.51	0.1-4.0	0.52
Second opinion x Sex			0.22
- No/not mentioned x Male	Reference		
- Yes, on the patient's initiative x Male	0.56	0.1-2.8	0.48
- Yes, on the health care provider's initiative x Male	0.10	0.0-1.4	0.18
Experienced delay x Sex			0.38
- No delay x Male	Reference		
- Not (clearly) mentioned x Male	0.89	0.1-5.8	0.90
- Yes, caused by the patient x Male	0.41	0.0-9.6	0.58
- Yes, caused by a health care provider x Male	0.79	0.2-3.2	0.74
- Yes, unknown who caused it x Male	9.25	0.7-125.8	0.10

*Statistically significant, P < 0.05
[a]Excluding GP and neurologist
[b]Excluding second opinion, initiative unknown
x Sex: interaction term of sex with variable (male = reference group)

Number of health care providers involved[a] x Sex is significant, meaning that there is a significant difference between men and women for the relationship between patient dissatisfaction and the number of health care providers involved.

In order to calculate the odds ratios for dissatisfaction with the involvement of 0, 1, 2 or ≥3 health care providers for women compared to men, we subsequently used a reduced model, excluding the non-significant interaction terms of sex with level of education, duration of the diagnostic pathway, second opinion and experienced delay.

Variable (n = 856)[b]	**Odds ratio (OR) for dissatisfaction**	**95% Confidence Interval (CI)**	**P-value**
Sex			<0.01*
- Male	Reference		
- Female	3.11	1.4-7.0	
Level of education			0.01*
- Low	Reference		
- Medium	0.41	0.2-0.8	<0.01*
- High	0.44	0.2-0.8	0.01*
Duration of the diagnostic pathway			0.44
- Unknown	Reference		
- <2 years	1.19	0.6-2.4	0.63
- ≥ 2 years	1.45	0.8-2.6	0.20
Number of health care providers involved[a]			<0.01*
- 0	Reference		
- 1	3.59	1.6-8.1	<0.01*
- 2	3.39	1.3-8.9	0.01*
- ≥3	4.91	1.3-19.1	0.02*
Second opinion			<0.001*
- No/not mentioned	Reference		
- Yes, on the patient's initiative	5.26	2.5-11.2	<0.001*
- Yes, on the health care provider's initiative	2.03	0.8-5.4	0.16
Experienced delay			<0.001*
- No delay	Reference		
- Not (clearly) mentioned	1.98	0.8-5.0	0.15
- Yes, caused by the patient	0.89	0.2-4.4	0.88
- Yes, caused by a health care provider	42.18	21.4-83.2	<0.001*
- Yes, unknown who caused it	7.20	2.7-19.3	<0.001*
Number of health care providers involved[a] x Sex			0.05
- 0 x Male	Reference		
- 1 x Male	0.19	0.1-0.6	<0.01*
- 2 x Male	0.52	0.1-2.4	0.40
- ≥3 x Male	0.62	0.1-4.5	0.64

*Statistically significant, P < 0.05
[a]Excluding GP and neurologist
[b]Excluding second opinion, initiative unknown
x Sex: interaction term of sex with variable (male = reference group)

Using this reduced model we calculated the odds ratio (OR) for dissatisfaction per number of involved health care providers, female compared to male.

Number of health care providers involved[a]	Odds ratio (OR) for dissatisfaction, female compared to male	95% Confidence Interval (CI)	P-value
- 0	3.11	1.4-7.0	<0.01*
- 1	0.58	0.2-1.4	0.23
- 2	1.63	0.4-5.8	0.45
- ≥3	1.92	0.3-12.1	0.49

[a]Excluding GP and neurologist

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4

Prodromal symptoms and early detection of Parkinson's disease in general practice: a nested case-control study

The most important role is to recognize it. The start of Parkinson's is often very vague and it is important for a GP to recognize it in time. (GP, male)

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Abstract

Background: Timely diagnosis of Parkinson's disease (PD), facilitating early intervention, depends largely on the GP's awareness of early symptomatology. For general practice, it is unknown which prodromal symptoms (symptoms preceding the typical motor symptoms of PD) demand the GP's alertness.

Objective: To assess prodromal symptoms that should alert the GP to the possibility of PD in primary care patients.

Methods: A nested case-control study was caried out in a population of approximately 12 000 patients registered in the Continuous Morbidity Registration database affiliated with the University of Nijmegen in the Netherlands. The database pools subject data from four primary care practices. The subjects comprised all 86 patients diagnosed with PD between 1972 and 2007, and 78 controls, matched by sex, age, socioeconomic status and primary care practice. The primary measures of outcome were the prodromal symptoms presenting in the 2 years prior to the diagnosis of PD. The number (and type) of referrals and diagnostic tests were also assessed.

Results: In the 2-year period prior to diagnosis, PD patients more often presented with functional somatic symptoms, constipation, hyperhidrosis and sleep disorders than controls. Patients also more frequently experienced more than one prodromal symptom and were more often referred within the primary care team or to a medical specialist.

Conclusion: Prodromal symptoms of PD are encountered in general practice. GPs should be alert when patients present with multiple prodromal symptoms in a 2-year period, especially considering the benefits of early intervention, and the future possibilities for disease-modifying therapy.

Background

Parkinson's disease (PD) is a common neurodegenerative disorder that has a severe impact on the patient's daily life.[1] Multiple caregivers are involved in the course of the disease.[2] The diagnosis of PD requires the presence of bradykinesia, combined with other motor symptoms such as muscular rigidity, a 4-6 Hz resting tremor or postural instability (UK Brain Bank Clinical Diagnostic Criteria).[3] However, it is becoming clear that non-motor symptoms such as autonomic dysfunction, depression, anxiety and cognitive decline are also part of the PD symptom complex.[4-6] Moreover, some motor and non-motor symptoms, including olfactory dysfunction and rapid eye movement (REM) sleep behavior disorder are prodromal symptoms; symptoms that are already present before the onset of the typical motor signs of PD.[4-7] Patients seem to be hindered by individual prodromal symptoms or combinations of them[8, 9] as they consult GPs and medical specialists with increasing frequency in the 10-year period before they are diagnosed with PD.[9]

Recognition of prodromal symptoms and increase in consultation frequency are essential for timely referral and early intervention, aiming to maintain the best quality of life for patients with PD.[3, 10-12] GPs play an important role in this, as patients will generally first present their symptoms to their GP, and not, for example, to a neurologist. Knowledge and awareness of the prodromal symptoms of PD are therefore crucial for GPs.[6]

Earlier studies of prodromal symtoms are mainly hospital-based and focus on patients referred to neurologists[4, 5, 7, 13]; only few studies are performed focussing on the population typically presented to a GP.[9] An Australian study showed there are deficits in the GP's knowledge of motor and non-motor aspects of PD. Knowledge of the prodromal symptoms of PD was not assessed.[14] However, given the low prevalence of PD in individual family practices, it seems likely that there are also deficiencies in the GP's knowledge of the prodromal symptoms.

This study therefore aims to characterize the prodromal symptoms of PD presenting in general practice and to give insight into referral rates in the 2 years prior to the diagnosis, in order to increase the GP's alertness for symptoms of PD.

Methods

Continuous Morbidity Registration database

We conducted a nested case-control study using data from the Continuous Morbidity Registration (CMR) database affiliated with the University of Nijmegen in the Netherlands. The recording in the database is anchored in the Dutch health care system where all citizens are registered with a personal GP, whether they consult the GP or not. Since 1971, all health problems are monitored in a population of approximately 12 000 patients from four general practices, representative of the Dutch population with regard to age and sex. In addition to health problems, sex, age, socioeconomic status (SES; low, middle and high) and marital status are registered.[15, 16]

All episodes of illness seen by or reported to the GP are registered as soon as they are established, using an adapted version of the E-list.[17] Monthly meetings are held with all participating GPs to discuss classification problems, monitor the application of diagnostic criteria and discuss coding problems in real and hypothetical cases. When necessary, diagnoses and codes are corrected. The validity of the CMR has been established repeatedly[15] and more than 65 papers based on CMR data have been published in international peer-reviewed journals between 1992 and 2012.[16]

Patients with PD

We selected all patients from the CMR database who were diagnosed with PD between 1972 and 2007. From 1972 to 1980, PD was diagnosed by a GP and from 1980 to 2007, by a neurologist. Patients must have been registered with the general practice for at least 2 years before their diagnosis. For a 2-year period prior to the diagnosis, the following variables were collected: sociodemographic characteristics; data on four major comorbid conditions (diabetes mellitus, cardiovascular diseases, COPD, rheumatoid arthritis); prodromal symptoms; and data on referrals and diagnostic tests. Based on a literature study, a selection of prodromal symptoms was studied: functional somatic symptoms; (autonomic) dysfunction, such as constipation, hyperhidrosis, olfactory dysfunction, orthostatic hypotension, urinary incontinence, swallowing difficulty, and sleep disorders; musculoskeletal complaints; neuropsychiatric disorders, such as anxiety, dementia, and depression; and traumata, such as fractures, laxations and distorsions.[4, 5, 7-9, 13] Functional somatic symptoms were defined as physical symptoms, appearing in patients with presumed psychosocial problems or psychological distress, that remain medically unexplained after adequate examination.[18]

Referrals were divided into three categories: somatic within the primary care team (physical therapist, social worker, occupational health officer, speech therapist, dietician and district nurse), somatic medical (all medical specialists except for a psychiatrist), and mental health (psychiatrist, psychologist and ambulatory mental health care). Diagnostic tests included hematological tests, X-ray examinations and ultrasonography.

Data on prodromal symptoms were obtained directly from the medical records, except for data on clear diagnoses and information about referrals and diagnostic tests. These data were derived from the CMR database. A patient was assumed to experience a prodromal symptom when a note was made in the medical record or when a diagnosis was coded in the database.

Controls

For each patient with PD, a matched control was drawn from the CMR population. The control matched the patient in sex, age, SES and primary care practice at the date the patient was diagnosed with PD. Furthermore, the match must have been registered at the same practice as the patient for a minimum of 2 years. The only exclusion criterion in the control

group was the diagnosis of PD. For controls, the same information as described for patients with PD was obtained.

Statistical methods

Statistical analyses were conducted using SPSS 20. Descriptive statistics were calculated. The chi-square test was used to assess the relationship between PD and the presence of the selected comorbid conditions. Conditional logistic regression was used in this nested case-control study to investigate the relationship between PD and the prodromal symptoms. The criteria for the matched sets were sex, age category (≤69 years, 70 to 80 years, ≥81 years), SES and practice. The numbers of cases and controls in the matched sets were uneven. In case a prodromal symptom did not present in the control group, a Fisher's exact test was used. In order to reveal patterns in the combined presentation of prodromal symptoms, all presented combinations of prodromal symptoms were scored. The chi-square test was used to assess the relationship between PD and the number of prodromal symptoms presented. Conditional logistic regression was used to investigate the relationship between PD and the number of referrals and diagnostic tests. The criteria for the matched sets were sex, age category (≤69 years, 70 to 80 years, ≥81 years), SES and practice. The number of referrals and diagnostic tests were divided into categories of frequency (0, 1, ≥2). There were varying numbers of cases and controls in the matched sets. The chi-square test was used to explore the relationship between PD and the number of referrals and diagnostic tests per individual (categories: 0, 1, ≥2).

A P-value less than 0.05 was considered statistically significant.

Results

Characteristics of subjects

We included 86 consecutive patients with PD and 78 controls (Table 1). There were no suitable controls for eight patients. The patient group consisted of 57% men, the mean age was 72.3 years (SD 8.8). Patients were of low, medium and high SES in 44.2%, 50.0% and 5.8% of the cases, respectively. Significantly more PD patients than controls had a cardiovascular disease (P 0.018; Table 1).

Characteristics	Patients (n = 86)	Controls (n = 78)	P-value
Sex (% male)	49 (57.0%)	42 (53.8%)	
Mean age (years)	72.3 (SD 8.8)	70.6 (SD 8.9)	
Socioeconomic status			
- Low, n (%)	38 (44.2%)	34 (43.6%)	
- Middle, n (%)	43 (50.0%)	39 (50.0%)	
- High, n (%)	5 (5.8%)	5 (6.4%)	
Comorbidity			
- Diabetes mellitus, n (%)	8 (9.3%)	4 (5.1%)	0.305
- Cardiovascular diseases, n (%)	36 (41.9%)	19 (24.4%)	0.018*
- COPD, n (%)	12 (14.0%)	9 (11.5%)	0.644
- Rheumatoid arthritis, n (%)	1 (1.2%)	1 (1.3%)	0.945

*P < 0.05, statistically significant.

Table 1. Characteristics of patients with Parkinson's disease and matched controls

Prodromal symptoms presented to the GP

Several symptoms presented by PD patients to the GP with higher frequency in the 2 years prior to the diagnosis, compared to matched controls (Table 2). There was a significant difference in functional somatic symptoms (OR 2.45; P 0.014), constipation (OR 3.32; P 0.039) and sleep disorders (OR 6.98; P 0.002) presented. Hyperhidrosis was only reported in the patient group (9.3% versus 0%; P 0.007). For all other symptoms, including musculoskeletal complaints, there was no significant difference between the PD patients and the controls.

Symptoms	Patients (n = 86)	Controls (n = 78)	Odds ratio (OR)	95% CI	P-value
Functional somatic symptoms	33 (38.4%)	17 (21.8%)	2.45	1.2-5.1	0.014*
(Autonomic) dysfunction:					
- Constipation	16 (18.6%)	4 (5.1%)	3.32	1.1-10.4	0.039*
- Hyperhidrosis	8 (9.3%)	0	-	-	0.007*[a]
- Olfactory dysfunction	1 (1.2%)	0	-	-	1.000[a]
- Orthostatic hypotension	2 (2.3%)	0	-	-	0.498[a]
- Urinary incontinence	9 (10.5%)	4 (5.1%)	3.21	0.8-13.2	0.106
- Swallowing difficulty	4 (4.7%)	1 (1.3%)	2.23	0.2-22.2	0.493
- Sleep disorders	22 (25.6%)	3 (3.9%)	6.98	2.0-24.3	0.002*
Musculoskeletal complaints	51 (59.3%)	37 (47.4%)	1.60	0.8-3.2	0.171
Neuropsychiatric disorders:					
- Anxiety	0	0	-	-	-
- Dementia	2 (2.3%)	2 (2.6%)	0.65	0.1-5.3	0.688
- Depression	3 (3.5%)	0	-	-	0.247[a]
Traumata	3 (3.5%)	7(9.0%)	0.33	0.1-1.4	0.123

[a] P-value Fisher's exact test

*P < 0.05, statistically significant

Table 2. Symptoms presented to the GP in the 2 years prior to the diagnosis Parkinson's disease; patients compared to matched controls

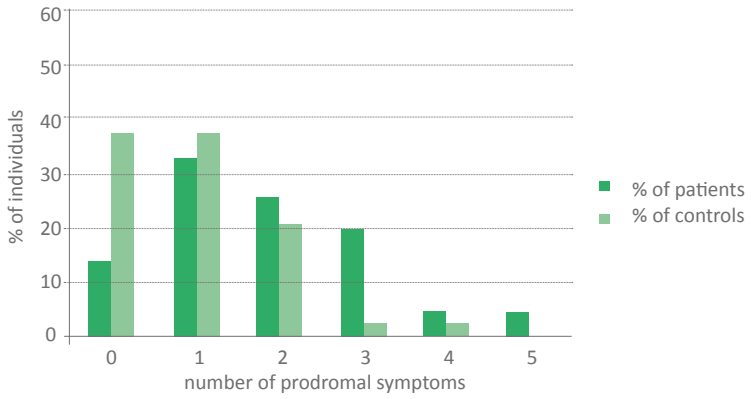

Figure 1. Number of prodromal symptoms presented to the GP in the 2 years prior to the diagnosis Parkinson's disease; patients compared to matched controls

Of all the PD patients, 53.4% presented with multiple prodromal symptoms in the 2-year period prior to the diagnosis, versus 25.6% of the controls (Figure 1) (OR 3.37; P < 0.001). We did not find any salient patterns in the combined presentation of prodromal symptoms (results not shown).

Referrals and diagnostic tests

Patients were referred more frequently than controls (Table 3). There was a significant difference in referral within the primary care team (OR 3.28; P 0.007). There was also a significant difference in referrals to a medical specialist; 46.5% of the PD patients were referred to a medical specialist at least once (OR 4.26; P 0.002), compared to 20.5% of the controls (OR 2.69; P 0.088). Referral to a psychologist or psychiatrist was very low.
More diagnostic tests were performed in the patient group than in the control group, but the results were not statistically significant.

Referrals/diagnostic tests	Patients (n = 86)	Controls (n = 78)	Odds ratio (OR)	95% CI	P-value
Somatic within the primary care team					
- 0	59 (68.6%)	69 (88.5%)			
- 1	23 (26.7%)	9 (11.5%)	3.28	1.4-7.8	0.007*
- ≥2	4 (4.7%)	0	-	-	-
Somatic medical					
- 0	46 (53.5%)	62 (79.5%)			
- 1	26 (30.2%)	10 (12.8%)	4.26	1.7-10.9	0.002*
- ≥2	14 (16.3%)	6 (7.7%)	2.69	0.9-8.4	0.088
Mental health					
- 0	77 (98.8%)	85 (98.7%)			
- 1	1 (1.2%)	1 (1.3%)	-	-	-
- ≥2	0	0	-	-	-
Diagnostic tests					
- 0	37 (43.0%)	46 (59.0%)			
- 1	30 (34.9%)	19 (24.4%)	1.94	0.9-4.0	0.077
- ≥2	19 (22.1%)	13 (16.7%)	1.99	0.8-4.8	0.126

*P < 0.05, statistically significant

Table 3. Number of individuals with 0, 1 or ≥2 referrals or diagnostic tests in the 2 years prior to the diagnosis Parkinson's disease; patients compared to matched controls.

Conclusions

Summary of the main findings

We identified that in primary care, PD patients more frequently experience prodromal symptoms in the 2 years prior to the diagnosis compared to controls. These prodromal symptoms include functional somatic symptoms, constipation, hyperhidrosis and sleep disorders. Over 50% of the PD patients presented more than one prodromal symptom. However, specific combinations of presented prodromal symptoms could not be found.
Referrals within the primary care team and to medical specialists occured more often in the patient group, whereas referral for mental health was low and comparable to controls. Diagnostic tests were requested equally for PD patients and controls.

Comparison with existing literature

We found that several prodromal symptoms, corresponding with those reported in the literature, were presented to the GP. Studies have shown that autonomic dysfunction and REM sleep behavior disorder can be present in the prodromal phase of PD.[4, 5, 8, 9, 19] Although we did not specify the nature of the sleeping disorders studied, it is likely that these included REM sleep behavior disorder.

Functional somatic symptoms diagnosed in the prodromal phase of PD might in fact be early autonomic symptoms of PD that are misdiagnosed, interpreted and registered by GPs as functional somatic symptoms. However, a recent study suggested a link between the pathophysiology of PD and a higher susceptibility of PD patients to functional somatic symptoms in the course of the disease.[20] In our study, functional somatic symptoms were recorded more frequently in PD patients than in controls. It is therefore possible that the higher susceptibility to functional somatic symptoms is already present in the years before PD is diagnosed. Regardless of the reason, GPs should be aware that the presentation of functional somatic symptoms could in fact be the presentation of the prodromal phase of PD.

In contrast to earlier studies [8, 9,] we did not find a significant difference in musculoskeletal symptoms between PD patients and controls. Since these symptoms already have high prevalence in the age group in which PD is mostly diagnosed, they seem to be of little value for GPs as distinctive signs of early PD.

Although the prodromal symptoms studied may not individually be specific enough to indicate early PD[6], our results show that two or more prodromal symptoms presenting in a 2-year period could be an indicator of early PD.

Our results demostrating the increased number of referrals in the patient group support results from earlier research.[9, 11] One study suggested that the increase in referrals reflects the variable non-motor symptoms that may present early or precede the motor phase of

PD.[11] Our study is in line with this, in that it showed that the increase in referral rate is not solely due to referrals to a neurologist.

Strengths and limitations of the study

The strength of this study is the use of a well-documented database (eliminating the risk of recall bias) to study the symptoms presented to the GP, the referrals and diagnostic tests in a 2-year period before the diagnosis of PD. Another strength is the inclusion of unselected cases of PD, in the early stage of their disease development. The database made it possible to recruit controls from the general population and, as a consequence, the findings hold external validity for PD diagnosis in primary care.

However, some methodological issues merit consideration. First of all, as for every longitudinal research, the number of patients included and the duration of the observation period are inversely proportional to each other. We chose a study period of 2 years prior to the diagnosis of PD, since for this period information could be retrieved for a substantial number of patients and controls. It cannot be excluded that a longer study period would lead to different results.

Secondly, we used a pre-selected list of prodromal symptoms, based on literature[4, 5, 7-9, 13] and supported by expert opinion. Because the main focus of our research was to extend the knowledge of GPs in order to increase their alertness, we studied known prodromal symptoms that were presented in general practice. The pre-selected list of prodromal symptoms was not intended to be comprehensive. It is therefore possible that some symptoms that occur in the early phase of PD, known or unknown at the time, were not included. An interesting target for future research could be identifying novel prodromal symptoms of PD by studying all symptoms presented in general practice by patients later diagnosed with PD.

A third point of attention is that only symptoms registered in the patient's medical record were included in this study. The results may therefore be an underestimation of the actual prevalence of prodromal symptoms, due to lack of presentation by the patient or lack of reporting by the GP. However, relying on symptoms captured in the medical health record of the patient closely resembles everyday clinical practice. Furthermore, by focussing on these symptoms, the analysis was based on symptoms significant enough for patients to consult and seek help from their GP.

Fourthly, attention needs to be given to the age categories designated for the conditional logistic regression. The age group of ≤69 years may seem too heterogeneous to form one group. However, this group consisted of a range of PD patients aged 52-69 years, with one outlier of 39 years. Since matching was also done by age, the distribution of age in the control group is comparable to the patient group. We therefore believe this heterogeneity has no noteworthy influence on the results.

A fifth point of attention is the possibility of confounding factors. The difference in the number of referrals might be partly explained by the difference in comorbidity between the group of PD patients and the control group. The presentation of adverse effects of medication could, in some cases, be similar with the presentation of some of the prodromal symptoms studied. Smoking habits and alcohol consumption could have also influenced the results.

Finally, our study did not comprehensively assess the prodromal symptoms of PD in general practice. It is therefore not possible to calculate the predictive values of these symptoms. However, the Dutch CMR database, with its large sample size, longitudinal design and proven validity [15] provided an important insight into the early phase of PD, suggesting that the first disabling symptoms of the disorder can be seen in general practice. Given the relatively low prevalence of PD in general practices, the results of this study can offer valuable support in handling the uncertainty around diagnosis, as is the case for other low-prevalence diseases.[21]

In conclusion

Prodromal symptoms are a reality in general practice. The therapeutic benefit of early intervention[3, 12] and the future possibilities for disease-modifying therapies, emphasize the importance of recognizing prodromal symptoms of PD leading to early diagnosis. This falls into the primary care domain and the role of GPs: awareness that functional somatic symptoms, constipation, hyperhidrosis, sleep disorders, and an increase in referrals may signal PD, leading to an early diagnosis.

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5

Transitions in Parkinson's disease in primary care: protocol of a longitudinal mixed methods study

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Abstract

Introduction: Parkinson's disease (PD) affects many aspects of the lives of patients and relatives. Patients must adapt continuously to disabilities that necessitate changes in (medical) support, such as domestic adjustments, involvement of (non) professional caregivers or admission to hospital. Such changes mark a transition: a transfer of a patient between levels or locations of care. Transitions are likely to be multifold and complex, given that PD care extends across all echelons of health care. Patients and relatives are vulnerable during a transition, which imposes risks for their safety and quality of life. Guidance by the general practitioner (GP), who knows the preferences of the patient, can help to overcome challenges associated with a transition. However, patient-centered primary care requires insight into the transitions PD patients encounter. We aim to examine these transitions and the way patients, relatives and GPs experience them and cope with them. Moreover, we will study the patients' expectations of their GP during a transition and the GPs' views on their role.

Methods and analysis: A longitudinal mixed methods study will be conducted, using qualitative research methods combined with quantitative data as a validated questionnaire on quality of life. Patients will be asked to make a video diary every 2 weeks for a period of 1 year. Once they encounter a transition, patients and their GPs will be interviewed to identify causes and consequences of the transition. The verbatim transcripts of the videos and interviews will be analyzed according to the principles of constant comparative analysis.

Ethics and dissemination: Ethical approval was not needed according to Dutch legislation. Informed consent of patients, relatives and GPs will be obtained. We will disseminate the results in peer-reviewed journals, at research conferences and on the website of the Dutch Parkinson's Disease Association.

Introduction

Parkinson's disease (PD) is a neurodegenerative, disabling disease characterized by motor symptoms and a wide variety of non-motor symptoms.[1] It affects physical, emotional and psychosocial aspects of the lives of patients and their relatives.[2, 3] The clinical presentation and rate of progression of PD vary considerably among patients[4, 5], as does the perception of the most troublesome problems.[6, 7] The complexity of the disease requires a multidisciplinary approach with active participation of the patient.[8] However, due to progression of PD and daily fluctuations of the symptoms, patients and relatives are forced to adapt continuously to new disabilities and limitations in daily life.[3, 9] Some of these disabilities and limitations necessitate changes in the support or medical care that is offered to a patient. Such changes mark a transition: a transfer of a patient between different levels of (non) professional care within the same location or between different locations of care.[10] Commonly encountered transitions are the need for domestic adjustments or specific tools for the patient, modification of pharmacotherapy, alternation in the involvement of (non) professional caregivers, adaptation of working hours or type of work and/or admission to specialized day care or hospital. Transitions are likely to be multifold and complex, given that PD care typically extends across all echelons of health care.[9, 11] They pose challenges to the communication skills and coping competencies of patients and professionals, since transitions emphasize the need for clarity in the preferences, expectations and roles of everyone involved. Moreover, transfers between different locations of care jeopardize continuity of care.[12, 13] Patients and relatives are particularly vulnerable and might feel overwhelmed by a transition, that is often unforeseen.[14] Safety and quality of life of patients and relatives are at risk.[12, 13]

The challenges associated with a transition can be partly overcome by a health care professional who is well aware of the care preferences of patients and relatives and who could guide them during a transition, if patients and relatives feel this need.[12, 15] In the Netherlands, the general practitioner (GP) is the preferred health care professional to fulfill this role as all patients are registered with a general practice; the GP coordinates access to specialized care[16]. This structure supports Dutch GPs to function as a family doctor, with insight into the physical and mental state of all family members and the contextual factors that influence the well-being. Furthermore, the GP has a long-term professional relationship with the patients and their relatives. No more than 20% of all patients with PD in the Netherlands are admitted to a nursing home somewhere in the course of their disease.[17] Over time, the vast majority of patients will, therefore, consult their GP with all sorts of health questions, thereby providing several occasions to discuss the expectations and preferences of the patient and the relatives.[18] Earlier research in the Netherlands showed that patients with a chronic disease (such as PD) appreciate a long-term relationship with their GP and his/her coordinating role.[19] Moreover, GPs have been shown to be aware of the need for customized, preferably proactive, care for chronically ill patients.[15]

Such proactive patient-centered primary care requires more insight into the transitions that patients with PD and their relatives encounter during the course of the disease. Therefore, this study aims to answer the following questions:

1. What transitions do patients with PD encounter? How do patients, relatives and GPs experience and cope with these transitions and what is the impact on patients' lives?
2. What are signs and symptoms of an upcoming transition?
3. What do patients with PD expect from their GP during a transition? Do GPs agree with these expectations?

Methods and analysis

Study design

A longitudinal mixed methods study[20] will be performed, using qualitative research methods as video diaries and in-depth interviews and quantitative data as a validated questionnaire on quality of life and ratings for the neurological signs of PD.

Participants

General practitioners

A purposive sample of general practices surrounding Nijmegen, the Netherlands, will be approached to participate. Purposive sampling will be used to increase the external validity and to provide a wide range of opinions. Based on expert experience and literature, we consider the following characteristics to be relevant for purposive sampling: geographical location of the practice (city vs. rural area); practice organization (group practice vs. solo practice); age, gender and working experience of the GP. GPs will be approached to participate until saturation in data analysis is reached.

Patients

The patient population will consist of patients with PD. In order to participate, patients need to meet the following inclusion criteria:

- The diagnosis of PD is confirmed by a neurologist, according to established guidelines.[21]
- The patient lives independently, possibly with help from (non) professional caregivers.
- The patient does not have a form of cognitive dysfunction (according to the GP) and is therefore, mentally capable of remembering what happened in the past weeks/months.
- The patient is capable of handling a simple video camera with clear instructions (possibly with help of a partner or significant other).

For each participating patient, the partner or a significant other will be asked to participate as well. Inclusion of patients (preferably with diversity in gender, age and Hoehn-Yahr stage of PD), data collection and data analysis will continue iteratively until saturation is reached.

Recruitment

General practitioners

GPs will be personally approached to participate. Subsequently, an email will be sent in which the aim of the study is explained briefly and more information is given to inform the GP what participation means for the GP, for the patient and for the partner/significant other. Once a GP confirms to participate, he/she will search the computerized patient files of the general practice to identify patients with PD that fit the inclusion criteria.

Patients

Patients that fit the inclusion criteria described above will initially be approached by their GP. The GP will give a brief explanation of the study and will ask if the patient is willing to be approached by the researcher. If so, the GP will inform the researcher and the researcher will contact the patient in order to send an information letter. A week later, the researcher will approach the patient again and will ask for questions regarding the study. When a patient (and possibly the partner/significant other) agrees to participate, an appointment will be made with the researcher in order to give more detailed information on the study and to explain the use of the camera. Moreover, this appointment will serve to ask the patient (and possibly the partner/significant other) for informed consent. Patients consenting participation can be included even when their partner/significant other does not consent to participate.

Data collection

Video diaries

Patients will be asked to make a video message of 5-10 minutes every 2 weeks for a period of 1 year. A pilot study already proved the feasibility of this method. Patients will use a basic video camera (JVC Pics 10, Sony MHS-FS1 or Panasonic HC-V110) that is provided to them for the purpose of this study. Patients will make the videos themselves in their own home, supported by an instruction manual. This manual includes an instruction on the use of the video camera and instructions regarding the content of every video message. A video message has to contain the following items:

- The name of the patient and the date of the recording.
- A grade between 0 and 10 (0 being the worst imaginable, 10 the best), reflecting the way the patient felt in the 2 weeks before the recording.
- Two tests for neurological signs of PD:
  - Finger tapping test, executed to conform to '3.4 Finger tapping' of the Movement Disorder Society-sponsored revision of the Unified Parkinson's Disease Rating Scale (MDS-UPDRS)[22]: 40 times with the left hand and 40 times with the right hand.
  - Arising from the chair, executed to conform to '3.9 Arising from chair' of the MDS-UPDRS[22].

- A description of three situations, that happened in the 2 weeks before the recording:
  - A situation that went very well, despite the fact that the patient has PD. What was the reason this went well?
  - A situation that did not work out the way the patient expected it to because of PD. What was the reason for it?
  - A situation in which the patient got help because of PD. Why was help necessary? Was the help asked for or offered? How did the patient experience the need for help and how did the patient experience the help itself?

When the patient's partner/significant other has agreed to participate in the study as well, he/she will be asked for a subsequent video message (following the recording of the patient) containing:

- A description of a situation, that happened in the 2 weeks before the recording, in which the partner/significant other gave help to the patient with PD. Did he/she offer to help or did the patient ask for it? How did it feel to help? And in general, how does it feel to be a caregiver?

Once a month, a research assistant will collect the video diaries. The assistant and the patient will also fill in the PDQ39, (a validated Dutch version of) a questionnaire on the quality of life of patients with PD.[23-25] Furthermore, the research assistant will explore whether a transition has taken place in the last month, using a short questionnaire in which the patient will be asked if any of the transitions, which are the focus in this study, have taken place.

Identification of a transition

A transition is defined as the transfer of a patient between different levels of (non) professional care within the same location or between different locations of care.[10] For this study, we will focus on a number of specific transfers, given they occur as a consequence of PD:

- A change in the extent of domestic help that is provided.
- A change in the extent of help that is necessary for personal care.
- A domestic adjustment (such as a bracket on the toilet or shower).
- The purchase of a specific tool (such as a walker or adapted cutlery).
- A modification of pharmacotherapy.
- The involvement of a health care provider (including, eg, the physical therapist and speech therapist), who was not involved before.
- Consultation of the GP or medical specialist, if not part of a routine follow-up.
- Adaptation of working hours or type of work.
- Admission to specialized day care or hospital.

In-depth interviews

Once a patient encounters a transition, he/she will be contacted to participate in an in-depth interview. The interviewer will stimulate the patient to tell more about the transition and the way the patient handled it. A brief topic list will be made to guide the interviewer, possible

topics are the patient's view on forerunner signs and causes of the transition, the role of caregivers during the transition and the impact of the transition on the life of the patient and his/her partner. As data collection and analysis will proceed as an iterative process, relevant and new topics will be added to the topic list after a preliminary analysis of every interview. In this way, ideas and thoughts that emerge in primary stages of the analysis will be brought forward in subsequent interviews as the study proceeds, in order to reach a deeper understanding of the relevant topics and themes.

The patient's GP will be approached for an in-depth interview on the transition as well. A different topic list will be used for the interviews with GPs, focusing on forerunner signs and causes of the transition, the role of the GP and the patient's prognosis concerning transitions due to PD. This topic list will also be modified according to the iterative process described above. GPs will be asked verbally (recorded on tape) for informed consent prior to the interview.
All interviews will be recorded on tape for the purpose of transcription.

Data analysis

The researcher will watch all recorded videos. Once a transition has taken place, the researcher will review the last four videos of the patient involved for cues of an upcoming transition and for specific details to be discussed in the interviews. The reviewed videos will be transcribed verbatim for analysis, ensuring the anonymity of the patient. Speech, facial expression, finger tapping and arising from the chair will be scored according to the MDS-UPDRS points 3.1, 3.2, 3.4 and 3.9, respectively.[22] PDQ-39 will be scored using the sum score of all subscores.[25] These sum scores will be plotted in a graph, that expresses the sum score per patient per month and the moment a transition has taken place.
All recorded in-depth interviews will be transcribed verbatim anonymously for analysis as well.

Analysis

The transcripts of the video diaries and in-depth interviews will be entered into ATLAS.ti 7, a software program for detailed coding in qualitative data analysis. In order to refine and focus the interview topic guides, analysis of the videos and in-depth interviews will start the moment the first transition has taken place and the videos and interviews have been transcribed. The analysis will be according to the principles of constant comparative analysis.[26] Two researchers will read all transcripts several times to familiarize themselves with the data. They will independently apply codes to meaningful words and sentences in the transcripts. These codes will be discussed, seeking agreement for their content. In case of disagreement, the opinion of a third researcher will be sought. Codes will then be grouped into themes that represent important and relevant aspects of transitions, as formulated in the research questions. Themes will be used to refine the interview topic guides and to progressively focus and explore the data in-depth. Analysis will continue until saturation is reached.

Ethics and dissemination

Longitudinal research with chronically ill patients poses several ethical issues.[27] One of these concerns the consent to participate. Patients will be asked for their written consent before they are included in the study. However, unforeseen circumstances and progression of the disease might change a patient's opinion. Consent will therefore be verified verbally before each interview. Moreover, when the researcher or research assistant suspects diminishing enthusiasm to participate, this will be brought up and the patient will be reminded that participation is voluntarily and can be ended (preliminary) at any time.

Furthermore, continuity in staff is a key element of longitudinal research. Therefore, we aim to assign one researcher, who will be responsible for watching all videos and performing the in-depth interviews. Moreover, this researcher will be part of the analyzing team. We also aim to make sure that the same research assistant will visit the participating patients every month.

The results of this study will be disseminated in peer-reviewed journals and at research conferences. Results will also be published on the website and in the magazine of the Dutch Parkinson's Disease Association. Moreover, the patients, relatives and GPs participating in the study will be informed about the results.

Discussion

In this study we will focus on transitions in PD in patients living at home. We will explore the transitions that patients encounter in the course of their disease and examine the way patients, their partners/significant others and their GPs experience these transitions. Moreover, our results will facilitate the anticipation of upcoming transitions. Finally, this study will offer the opportunity to compare the patients' expectations of their GP during a transition and the GPs' views on their role in a transition. Therefore, this study will provide insight into crucial elements of transitions in the course of PD, such as the extent to which patients want their GP to be involved during a transition and how this could influence their experiences. Moreover, this could give insight into the most ideal moment to offer primary care during a transition. This will enable improvement of (proactive) primary care for patients with PD, in a patient-centered way.

Recruitment challenges

Recruiting patients with a chronic disease to participate in longitudinal qualitative research poses difficulties.[27] Furthermore, recruitment of GPs and patients in primary care can be challenging.[28] In this study we expect that the challenges at the GP's level include willingness to participate and to fulfill all requests associated with participation: searching the computerized patient files for patients that fit the inclusion criteria, approaching suitable patients, informing the researcher when a patient is willing to be approached, and participating in an in-depth interview.

Moreover, there are possible challenges at the level of the patient. The number of patients with PD in each general practice is about four[29] and only a selection of these patients will fit the inclusion criteria. One could also argue that the patients that do fit the inclusion criteria are relatively independent patients with PD. It is very well possible that this group of patients will encounter different transitions than patients with more advanced PD. However, the GP is the main caregiver for patients with PD who still live at home; therefore we specifically aim to study this relatively independent group. If the present approach is successful, future work should address the transitions in more advanced PD. Apart from this, patients might find it difficult to agree with participation for a period of 1 year, as they are uncertain what the next year will bring to them. Unforeseen circumstances could force the patient to stop his/her participation preliminary. Furthermore, participating patients might have difficulties in handling the video camera or describing useful situations in their video recordings. However, a pilot study suggested that patients with PD are able to make useful video recordings themselves by following the instruction manual provided to them.

Challenges at the level of the relative could also influence recruitment, such as the inability to handle an additional, non-essential, task besides care giving or their opinion that participation in a study might not be good for the patient.

Strengths and limitations

The home setting, in which the videos will be recorded, might comfort patients and partners to freely express symptoms and difficulties they experience in daily life, even those they might interpret as too unimportant to consult the GP for although these might have a significant impact on the quality of life.

The longitudinal qualitative design of this study provides other unique opportunities.[27] For example, it will be possible to interview patients and their GPs at turning points in life rather than at fixed moments. Furthermore, the information from the video diaries combined with the iterative process of data analysis enables refinement and customization of the topic guide for each interview. This will give a deeper understanding of the transitions encountered in the course of PD, and the causes and consequences of these over time. In addition, the results of the in-depth interviews with the patients and their GPs enables comparison between the patient's views and expectations and those of the GP, providing insight into possible discrepancies. Moreover, the video diaries allow for within-patient comparison of the results, since progression of the disease and the constant adaptation to new disabilities or limitations might influence the experiences and preferences patients express over time. Finally, the use of multiple qualitative research methods, supported by quantitative data, offers valuable opportunities for data triangulation.[20] The constant comparative content analysis, applied until saturation is reached, adds further to the robustness and validity of the results.[26]

However, qualitative research methods have disadvantages as well. Qualitative research has neither the goal nor is the suited method to quantify variables or to generalize results from a small sample to a larger population. Therefore, the results of this study will need to be interpreted in the light of the studied sample of patients and GPs, and the context and culture of the Dutch health care system.

Ethical approval

The research ethics committee of the Radboud university medical center was consulted and decided approval was not needed according to the Dutch legislation (correspondence date December 11th 2013). Patients, partners/significant others and GPs will be asked for informed consent.

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6

Being in control of Parkinson's disease: a qualitative study of patient experiences

I have the feeling that I'm less affected by it because I manage the situation. It's the feeling that you're the master of your own fate. (Male, 76 years, H&Y2)

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Submitted

Abstract

Background: Chronically ill patients will face and have to cope with changes in care. Research so far has focused on the experiences and coping of patients with prevalent diseases for which the general practitioner (GP) offers care. We lack knowledge of changes in care of patients with low-prevalent diseases such as Parkinson's disease (PD), for which care is usually offered by medical specialists.

Objectives: To improve knowledge of PD patients' experiences and coping with changes in care.

Methods: A qualitative interview study with a purposive sample of 16 community-dwelling PD patients in the Netherlands. Semi-structured interviews were analyzed using an inductive approach to comparative content analysis.

Results: Patients mentioned a variety of changes in care such as changes in the level of unpaid care, the purchase of tools, modification of pharmacotherapy or admission to hospital. Three themes had a great influence on patients' experiences and their acceptance of impairments afterwards: anticipating change; self-managing change; and managing expectations. Self-management was facilitated by patients' ability to anticipate change. Patients preferred to self-manage change as this gave them a feeling of control. Self-management was also related to realistic expectations and acceptance of the post-change situation.

Conclusion: Self-management support for PD patients can be offered by GPs, as the changes in care patients experience are likely to be the cumulated result of disease-related and patient-related factors GPs are familiar with.

Introduction

Chronically ill patients will face changes in care at some point in the course of their disease. Patients with Parkinson's disease (PD), for example, suffer from motor symptoms and a wide variety of non-motor symptoms, and the fluctuating expression and progression of this disease frequently force PD patients to adapt to new impairments and disabilities.[1] Some of these disabilities will necessitate changes in care.[2]

Changes in care may include changes in home help services, the purchase of a tool or modification of pharmacotherapy.[3] So far, however, research has mainly focused on transitional care from one health care setting to another or to home.[3-7] Studies among patients with prevalent chronic conditions such as heart failure, chronic obstructive pulmonary disease (COPD) and cancer show that patients feel they lack control when discharged from hospital. Moreover, they feel insecure, unprepared, inadequately guided and not involved in care-taking decisions at those moments.[6, 7]

Offering support to prepare patients to handle changes in care can contribute to patients' well-being.[8] The insight is growing that patients' health care needs to deal with the challenges of a chronic disease are defined not only by the disease itself, but also – and perhaps even more so – by patients' ability to cope.[9] As general practitioners (GPs) offer disease-specific care to patients with prevalent conditions such as COPD and diabetes mellitus and are familiar with patients' personal context and ability to cope, GPs are the most appropriate health care providers to offer medical and mental support to patients with COPD and diabetes experiencing changes in care.

In the case of a less prevalent disease such as PD, however, it is not clear who should be offering this support, as the neurologist offers disease-specific care, while the GP is familiar with patients' personal circumstances. In order to offer PD patients customized support in handling changes in care, we need to improve our understanding. In this study, therefore, we aim to increase our knowledge of community-dwelling PD patients' experiences and coping with the changes in care they face in the course of their disease.

Methods

Study design

We performed an exploratory qualitative study using semi-structured interviews with community-dwelling PD patients in the Netherlands. A qualitative research design and purposive sampling of patients were chosen to gain wide and in-depth knowledge of patient experiences and coping.

The research ethics committee of the Radboud university medical center examined the study protocol and concluded that the study could be carried out in the Netherlands without requiring approval by the accredited regional research ethics committee (December 11th 2013). Written informed consent was obtained from all participating patients.

Selection of study subjects

Between September 2013 and November 2014, nine GPs working in and around Nijmegen (The Netherlands) were asked to select patients that met the following criteria: a diagnosis of PD (established by a neurologist according to accepted criteria); community-dwelling; no apparent cognitive dysfunction; and capable of handling a video camera with instructions. A purposive sample of patients - based on age, gender and severity of PD according to Hoehn and Yahr (H&Y)[10] - was approached by their GP.

When a patient agreed to participate, the researcher AP (a medical doctor with experience in qualitative research) gave more detailed information and asked the patient for informed consent. Initially patients were asked to participate for a period of 1 year. After inclusion of eight patients, we reduced the study period to 6 months because patients mentioned that 1 year was rather too long. The final number of included patients was determined by the point of data saturation. Data collection ended in June 2015.

Data collection

Supported by an instruction manual, patients made a video once a fortnight to give a verbal and non-verbal impression of their physical and mental state.[11] Once a month, a research assistant collected these videos and explored whether a change in care was experienced. Special interest was paid to changes that may occur in the course of PD.[11]

If patients had experienced a change in care, they were interviewed face-to-face by researcher AP in their own home. A brief topic guide with core questions and optional prompts was available, which was customized using specific individual information from the patients' videos (Table 1). Interviews were recorded and fully transcribed.
More detailed information on recruitment and data collection can be found in the study protocol.[11]

Topic	Questions
A	Can you describe the change in care that took place?
B	Can you tell me about your experiences with this change in care? - How did you experience it? - Can you describe if anything or anyone had an influence on your experience? And if so, how did this influence your experience?
C	Can you tell me about your coping with this change in care? - How did you cope with this change in care? - Why did you cope with it this way? Can you describe your considerations? - Can you describe if anything or anyone had an influence on your coping? - And if so, how did this influence your coping?

Table 1. Semi-structured interview topic guide (original version)

Data analysis

ATLAS.ti 7, a computer program for qualitative data analysis, was used for coding. Analysis of the anonymous transcripts started as soon as the first interviews had been transcribed and was an iterative process using an inductive approach to comparative content analysis.[12, 13] Two researchers (AP, AvL) independently read all transcripts and applied codes to meaningful words or sentences. Codes were discussed, seeking agreement for their content. New codes arising from these discussions were applied to the transcripts. Codes were grouped into themes, and final themes were agreed upon with the supervisory committee (all authors). Themes were used to adapt the interview topic guide and to progressively focus and explore data in-depth. After analysis of 16 interviews with 12 different patients, no significant new codes emerged. Conduction and analysis of five additional interviews confirmed saturation.

Results

Study population

Sixteen patients participated in the study. Three patients did not complete the follow-up period: one patient died, and two patients withdrew because they experienced difficulty with storytelling to the video camera or the burden of comorbidity. Two participating patients did not experience any changes in care. Thirteen patients and one personal caregiver (replacing the patient who had died during the study period) were interviewed about a total of 34 changes in care. Some patients were interviewed more than once because they experienced changes at different moments. A total of 21 interviews were conducted, each taking between 60 and 90 minutes.

The patients' mean age at the start of the study was 68 years (SD 6.0) (Table 2). Most patients were male (n = 11). The majority had an H&Y stage of 2 or less (n = 12), and two patients had an H&Y stage of 4.

Experienced changes in care

Patients experienced a variety of changes in care such as changes in the level of unpaid care to prepare meals, the purchase of tools such as an adapted cup or the modification of PD-related pharmacotherapy because of hallucinations. Two patients were admitted to hospital acutely, and one patient experienced a planned admission for further investigation (Table 2).

Patient Code (A-P)	Sex	Age at start of study (years)	Severity of disease Mild-to-severe (H/Y stage 1-4)	Experienced change(s) in care (Each line represents a separate interview and shows the discussed changes in care)	Follow-up period (months)
A	Male	65	1	- Modification of PD-related pharmacotherapy - Domestic adjustment	12
B	Female	58	1.5	- Purchase of a tool - Change in unpaid care - Domestic adjustment	12
C	Male	59	1.5	- Change in unpaid care	12
D	Male	76	2	- Modification of PD-related pharmacotherapy (adverse effects)	12
E	Male	63	2	- Purchase of a tool - Domestic adjustment	12
F	Male	67	1.5	- Acute admission to hospital, modification of PD-related pharmacotherapy	12*
G	Male	65	1.5	- Involvement of a speech therapist - Consultation of GP, consultation of neurologist, modification of PD-related pharmacotherapy	12
H	Male	79	4	- Acute admission to hospital	12*†
I	Female	75	1	- Consultation of GP, planned admission to hospital - Consultation of neurologist, modification of PD-related pharmacotherapy	6
J	Male	73	2	- Domestic adjustment, modification of PD-related pharmacotherapy - Consultation of neurologist	6
K	Female	70	4	- Consultation of GP, consultation of neurologist, modification of PD-related pharmacotherapy	6
L	Male	72	2.5		6*
M	Female	70	1		6
N	Female	64	2.5	- Modification of PD-related pharmacotherapy	6
O	Male	65	1.5	- Consultation of GP, ENT specialist and neurologist, modification of pharmacotherapy (laxative)	6
P	Male	72	2	- Consultation of neurologist, consultation of GP, modification of pharmacotherapy (laxative)	6

*Did not finish follow-up, †Deceased during follow-up

Table 2. Characteristics of the participating patients with Parkinson's disease
- including an oversight of the experienced changes in care -

Themes influencing patients' experiences and coping

We identified three themes that had a major influence on patients' experiences, their coping with changes in care and their acceptance of post-change results: anticipating change; self-managing change; and managing expectations. These themes will be explored below and will be illustrated with quotations.

Anticipating change

Changes in care that were not foreseen by patients could be overwhelming for them. A patient who had been admitted to hospital acutely realized that admission was inevitable but felt that his wishes had not been attended to, which contributed to his experiencing lack of control at the moment of the change.

I had the idea that Parkinson's caused my complaints...I told him [the GP] 10 times: "That is what I believe."…but he persisted and then I had to go to hospital. (Male, 67 years, H&Y 1.5)

Patients who expected a change to occur were mostly able to anticipate it and to – for example – make domestic adjustments or purchase a tool. Patients would then feel they were in control and it was less complicated for them to accept any possible impairments that remained despite the change. Anticipating change was easier for patients if they had sufficient knowledge of the disease.

Well, it [encountering a change in care] isn't difficult. […] I know it's a progressive disease. […] I anticipate. (Male, 63 years, H&Y 2)

Self-managing change

Patients preferred to solve the problems they encountered themselves, for example by making a domestic adjustment or by searching for a proper balance between the therapeutic and adverse effects of PD-related pharmacotherapy. Loss of this ability to self-manage would lead to feelings of anger, grief and dependence.

Sometimes I'm angry that I'm no longer able to do it myself. […] At those moments, I've got the feeling I should still be able to do it. Yes, it makes me sad. (Female, 58 years, H&Y 1.5)

It's hard to be dependent. I'm not used to needing help. […] I prefer doing things myself. When you need help, it means you depend on other people. (Male, 65 years, H&Y 1)

Asking a relative to help was very difficult, and patients' experiences differed depending on the person they asked for help.

I think that's a matter of embarrassment again. What will she [my neighbor] think of me? Will she think that I can't do anything on my own anymore? [...] I don't have that with my husband, because he is so familiar. [...] When I ask my neighbor, I feel like I'm a whiner. (Female, 58 years, H&Y 1.5)

If patients were able to solve their problems independently, they considered themselves to be the manager of the change in care, which led to a sense of control over the change itself and the situation after the change.

Nowadays I take my medication more knowingly, taking into account that if I take my medication now, I'll have my 'low' in two hours' time. I have the feeling that I'm less affected by it because I manage the situation. […] It's the feeling that you're the master of your own fate. (Male, 76 years, H&Y 2)

If the help of a health care provider was inevitable, patients emphasized the importance of shared decision-making as this allowed them to still be involved and to maintain control over the change in care.

During the consultation with the neurologist we decided together to change the medication dose. (Female, 70 years, H&Y 4)

One patient, however, stressed that, if a health care provider was involved, she did not want to be the one making the decision.

I don't want to do that [make the decision]. […] In my opinion, when a health care provider [neurologist] tries to help you, you should just listen and do what he says. (Female, 75 years, H&Y 1)

Managing expectations

If a health care provider initiated a change in care, for example the modification of PD-related pharmacotherapy when symptoms got worse, patients had unrealistically high expectations of its results. As these expectations were usually not met, patients were disappointed and felt uncertain.

Yesterday I took the pills at 10 pm. At 11 pm my husband switched off the light. When I wanted to turn over in bed, I was barely able to do it. But I had just taken those pills! I should have been able to do that [turn over] by then, shouldn't I? (Female, 64 years, H&Y 2.5)

Moreover, they had difficulties accepting the impairments that remained after the change.

The effects I expected of the modification of my medication failed to happen...for example fewer dips or a shorter period; that I would experience less trouble due to dips. Actually, that

I'd just feel better. […] However, looking back at the past few weeks, I'm disappointed about the effect. (Male, 65 years, H&Y 1)

If patients came up with their own solutions to the problems they faced, however, they had realistic expectations of the change, which helped them accept the post-change situation, even if their impairments were not remedied.

Well, I wanted something [an electric shaver] that would be more comfortable, so I didn't have to be afraid of cutting myself with a razor. (Male, 63 years, H&Y 2)

Discussion

Main findings

Community-dwelling PD patients experience a variety of changes in care such as changes in the level of unpaid care, the purchase of tools, modification of pharmacotherapy or admission to hospital. Three themes influence patients' experiences and their acceptance of impairments remaining after change: anticipating change; self-managing change; and managing expectations. Being able to anticipate change enhances patients' self-management and feeling of control. Patients prefer to self-manage changes in care, and if they succeed in doing so, they have realistic expectations and can accept impairments that remain despite the change. If a change is initiated by a health care provider, however, expectations are often unrealistically high and unmet.

Strengths and limitations

To the best of our knowledge this is the first study that explores community-dwelling PD patients' experiences and coping with the changes in care in the course of their disease. The design of this study has several advantages. The monthly visits of the research assistant enabled us to respond quickly to changes in care and to interview patients shortly after these turning points in life, thereby limiting the risk of recall bias. Moreover, the interview prompts, based on patients' videos, contributed to questioning in-depth. All interviews were performed by the same skilled interviewer, who had no professional relationship with the patients and who was also part of the analyzing team. We feel confident that we were able to make a valid contribution to the knowledge of the experiences of most community-dwelling patients with PD and their coping with changes in care.[13]

However, some limitations need to be taken into account. Despite purposive sampling, we included more men and patients with mild-stage disease. The use of technical equipment and the long duration of data collection might have influenced this. The (expected) burden of making videos was a frequently mentioned reason not to participate or to withdraw early and it is possible that sampling bias occurred as a result of this; patients willing and able to use technical equipment – such as the patients included in this study - might for example be more inclined to self-manage. Our population, finally, came from a single regional setting in

the Netherlands with well-developed specialized PD care. Patients in different areas might have different care experiences.

Interpretation in relation to existing literature

Our study shows that community-dwelling PD patients frequently experience changes in care that occur in their personal context. These changes may be a direct result of PD alone, but they are more likely to be the cumulative effect of the disease and patient-related factors such as comorbidity, ageing, personal circumstances and the ability to adapt and cope. Domestic adjustments, for example, depend on a patient's living situation. Informal care, moreover, will only be offered if a patient has relatives who are willing to provide it. In addition, coping with the impairments of PD by buying a tool reflects a patient's ability to adapt.

Self-management is acknowledged to be an essential part of coping with a chronic disease[14], and it is frequently described as patients' competence to deal with the symptoms, treatment and consequences of a disease while maintaining quality of life.[15, 16] If people develop successful coping strategies to deal with the problems they face in their daily lives, their subjective well-being does not have to be affected by new impairments.[9, 17] Resilience is crucial in this. Huber et al. criticize the 1948 World Health Organization's (WHO) definition of health and propose to view health as 'the ability to adapt and to self-manage'.[17] Studies show that resilience and self-management can contribute to patients' well-being and experienced health.[14, 16] Self-management, furthermore, is known to empower chronically ill patients and to add to patients' self-efficacy, independence and feelings of control over life.[14, 16, 18] Our results are in line with this. If patients are able to self-manage the changes in care they face, they feel in control. The loss of this ability, on the other hand, leads to anger, grief and feelings of dependence.

Just like patients with multiple sclerosis (MS), PD patients are challenged in their self-management by the progressive nature of the disease and the fluctuating expression of symptoms.[2, 19, 20] Self-management is closely related to the ability to think ahead and plan changes, taking into account personal boundaries.[18, 19] MS patients mention they look for knowledge of the disease to be able to anticipate changes.[20] Our study contributes to this by showing that knowledge of potential changes in care is important as well. Patients who realize that their disease will progress and that changes in care are inevitable at some point are able to anticipate and even initiate changes at a moment and in a way that suits them. This leads to a feeling of control and helps them to have realistic expectations of changes in care and to accept possible remaining post-change impairments.

Implications for clinical practice

Health care providers can contribute to community-dwelling PD patients' well-being by stimulating and facilitating self-management and by providing information about the disease, treatment options and potential changes in care. As GPs offer most of the disease-specific

care, they are already experienced in providing self-management support to patients with common chronic conditions such as COPD and diabetes mellitus.[21, 22] Although PD is a less prevalent disease and disease-specific care is provided by specialized health care providers, GPs can still offer self-management support. After all, the changes in care that PD patients face are likely to be the cumulative effect of the disease itself and patient-related factors, which GPs are well acquainted with. Their knowledge of facilitating and complicating factors in their patients' situation, moreover, enables GPs to offer help in establishing realistic self-management goals.[23]

Conclusion

PD patients face a variety of changes in care they prefer to self-manage. If patients succeed in doing so, they have realistic expectations and can accept impairments that remain despite the change. Being able to anticipate a change enhances self-management. Expectations of changes initiated by health care providers are often unrealistically high and unmet.

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7

Parkinson's disease: patient and general practitioner perspectives on the role of primary care

The GP doesn't have a big role in the treatment of PD, but you have to be aware of what is happening, there's more to the patient than just PD. (GP, male)

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Abstract

Background: Specialized Parkinson's disease (PD) care offers advantages to patients. However, specialized health care providers may be unaware of patients' personal context and comorbidity, leading to conflicting treatment regimens. Patients may benefit from a more holistic approach.

Objective: To clarify the role community-dwelling PD patients see for general practitioners (GPs) in PD care and to clarify the role GPs see for themselves.

Methods: Qualitative interview study with 16 community-dwelling PD patients and 12 GPs in the Netherlands, using a constant comparative approach to analysis.

Results: Patients expressed a preference for self-management and autonomy in decision-making. GPs chose a limited, reactive position in early-stage PD care to stimulate patient autonomy. Moreover, GPs felt insufficiently competent to extend their role. Patients also felt GPs lack expert knowledge and skills; they focus on their neurologist for PD care. In addition, GPs observed patients might not realize what accessory role the GP could have, a role GPs described as essential in being aware of patient's well-being. Patients did not describe additional roles for the GP in more advanced disease, whereas GPs mentioned a shift towards a more proactive and extended role.

Conclusion: Patients and GPs see a limited role for the GP in early-stage PD care because of patient autonomy and GP's lack of specific knowledge and skills. However, GPs should feel more confident of the added value of their generalist approach to care for patients with a complex chronic disorder as PD. If generalist and specialized care reinforce each other, PD patients benefit.

Introduction

Parkinson's disease (PD) is a complex neurodegenerative disorder that varies considerably in clinical presentation and rate of progression.[1-3] The majority of all patients with PD live at home, where the general practitioner (GP) offers care. In the Netherlands, all citizens are registered with a personal GP, who has a central position in the health care system.[4] Amongst other tasks, GPs play an important role in chronic disease care. They are well trained in providing everyday care for common chronic conditions such as diabetes mellitus and chronic obstructive pulmonary disease.[5, 6] Knowledge of the patient's personal context facilitates and enhances the quality of this care.[7]

However, in case of PD and other complex chronic conditions – conditions with uncommon presentations or complicated treatment regimens – care is often best provided by specialized health care providers with expert knowledge and skills.[7, 8] On the other hand, patients with PD frequently suffer from more than one chronic condition.[9] Involvement of various specialized health care providers, each focused on one condition and unaware or unconsidered of the patient comorbidity, could lead to conflicting treatment regimens.[9, 10]

Moreover, community-dwelling PD patients encounter many care transitions in their home environment. These transfers in the amount or type of care that is offered include for example domestic adjustments or the purchase of tools.[11, 12] Such transitions and the impact on the patient context usually stay out of sight of specialized health care providers.

Community-dwelling PD patients may therefore benefit if care is not exclusively provided by specialized health care providers. For example, there may be a role for the GP in PD care as well. However, it is unknown whether PD patients and GPs themselves recognize a role for the GP in PD care. We therefore aim to clarify the role community-dwelling PD patients see for their GP in PD care. Moreover, we want to clarify the role GPs see for themselves.

Methods

Recruitment

Between September 2013 and June 2015, a longitudinal qualitative study was performed.[13] A purposive sample of GPs of nine general practices in and around Nijmegen – a city in the eastern part of the Netherlands – were asked to select patients who met the following criteria: a diagnosis of PD (established by a neurologist according to accepted criteria); community-dwelling; no apparent cognitive dysfunction; and capable of handling a video camera with instructions. A purposive sample of these patients, based on age, gender and severity of PD according to Hoehn and Yahr (H&Y)[14], were approached by their GP (Figure 1).

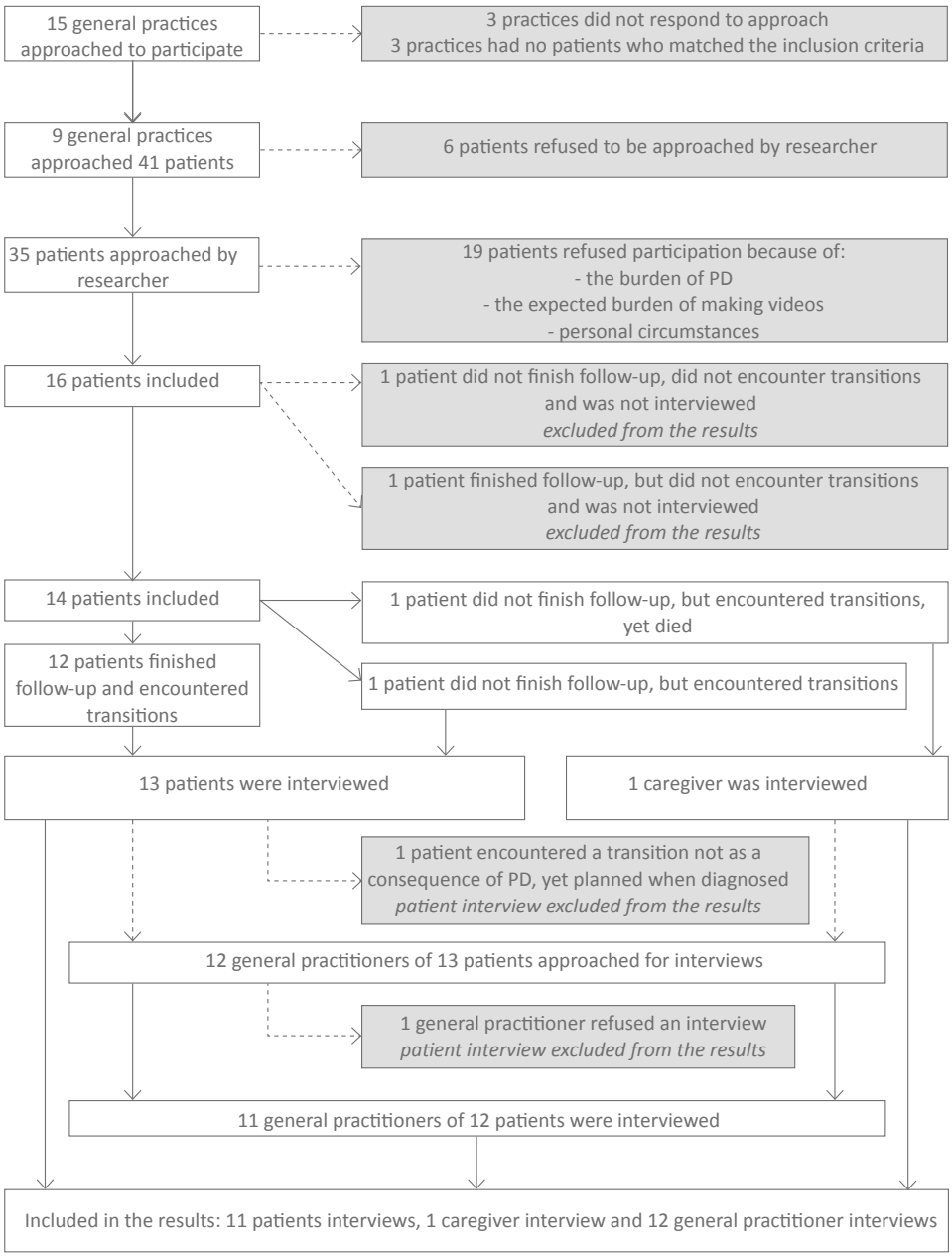

Figure 1. Flowchart from recruitment of patients with Parkinson's disease and their general practitioners to analysis

When a patient agreed to participate, the researcher (AP) gave more information and asked the patient for informed consent. Initially patients were asked to participate for a period of 1 year. After inclusion of six patients, we reduced the study period to 6 months because patients mentioned 1 year was rather long.

We chose a qualitative research design and purposive sampling of GPs (based on gender and location of the practice) and patients to gain wide and in-depth insight into the role patients and GPs see for the GP. We expected that inclusion of 15 patients and their GPs would lead to saturation.

Data collection

Patients made a video once a fortnight. Each video contained the same items: a grade between 0 and 10 (0 being the worst imaginable, 10 the best), reflecting the patient's feelings in the 2 weeks before the recording; two tests for neurological signs of PD; a description of a situation that went very well, a situation that did not work out as planned and a situation in which the patient needed help; and the possibility to add free comments.[13] Once a month, a research assistant collected the videos and explored whether a transition had taken place. The definition of a transition used in this study is the transfer of a patient between different levels of professional or non-professional care within the same location or between different locations of care.[11] We focused on a number of specific transfers that are likely to occur as a consequence of PD (Table 1).

Care transitions
- A change in the extent of domestic help that is provided
- A change in the extent of personal care help that is provided
- A domestic adjustment (such as a bracket on the toilet or shower)
- The purchase of a specific tool (such as a walker or adapted cutlery)
- A modification of pharmacotherapy
- The involvement of a health care provider (including for example the physical therapist and speech therapist), who was not involved before
- Consultation of the general practitioner or medical specialist, if not part of routine follow-up
- Adaptation of working hours or type of work
- Admission to specialized day care or hospital

Table 1. Care transitions as a consequence of Parkinson's disease, included in the study

Patients were questioned by the researcher (AP) in a semi-structured interview once they encountered a transition, rather than at predetermined moments. A brief topic guide was used that focused on the role of the GP in PD care (Table 2). Information from the videos was used to customize the topic guide and to provide prompts, for example by referring to the described situation in which the patient needed help or a patient's considerations regarding consultation of the GP. The patient's GP was asked for informed consent and interviewed by the researcher (AP) on the same subject (Table 2). All interviews were recorded on tape for the purpose of transcription.
We refer to the study protocol for more detailed information on recruitment and data collection.[13]

Interviewee	Topic	Questions
Patient/caregiver	A	What kind of change in care did you face?
	B	Which health care providers are involved in your care?
		Did you discuss the change with your general practitioner or neurologist?
		Why/Why not?
		What did you expect of them?
	C	What do you expect in general from the health care providers involved in PD care?
		What role do you see for each one of them?
General practitioner	A	Do you know what change in care your patient faced?
	B	Did you have a role in this change?
		Do you see a role for yourself?
		Do you feel that you were able to guide the patient in this change?
		Why/Why not?
	C	What role do you see in general for the GP in PD care?
		What do you think the patient expects from you?

Table 2. Topic guide for semi-structured patient and general practitioner interviews concerning the role of the general practitioner in Parkinson's disease care

Data analysis

All recorded interviews were transcribed verbatim and made anonymous for analysis. Software program ATLAS.ti 7 was used for detailed coding in qualitative data analysis. In order to refine and focus the interview topic guide, analysis of the interviews started as soon as the first interviews were transcribed. Data analysis was an iterative process using a constant comparative approach.[15] Two researchers (AP, CV) familiarized themselves with the transcripts and independently applied codes to meaningful words and sentences. Codes were discussed, seeking agreement for their content. Results of the analysis of interviews

of patients and GPs were compared. After analysis of 10 patient interviews and 8 GP interviews, no significant new codes emerged. Conduction and analysis of two additional patient interviews and four additional GP interviews confirmed saturation. Codes were then grouped into themes that were discussed and agreed upon by the supervisory committee (ToH, AL).

Results

A total of 35 patients from nine general practices were approached to participate, of whom 19 refused. Finally, 16 patients and 12 GPs participated. Three patients did not complete the follow-up period: one patient died, another was over burdened by comorbidity and the third patient had difficulties with storytelling to the camera (Figure 1).

Thirteen patients and the caregiver of the deceased patient were interviewed about 34 encountered transitions, varying from the purchase of a tool to hospital admission. GPs of 12 patients were interviewed as well: one GP was interviewed twice, concerning two different patients. Interviews lasted 30 to 90 minutes. Only those cases in which both patient and GP were interviewed were included in the results (n = 12 patients, n = 11 GPs; Table 3) (Figure 1).

The mean age of the included patients at the start of the study was 69 years (SD 6.0). Most patients were male (n = 8). The majority had mild stage disease (H&Y ≤ 2) (n = 10), two patients had an H&Y stage of 4.

Most participating GPs were male (n = 7) and worked in the city (n = 7). All GPs worked in a group practice (Table 3).

Patients' characteristics						General practitioners' characteristics	
Patient Code (I-XII)	**Sex**	**Age at start of study (years)**	**Severity of disease Mild-to-severe (H/Y stage 1-4)**	**Encountered transition(s)**	**Follow-up period (months)**	**Sex**	**Area**
I	Male	65	1	Modification of pharmacotherapy for PD	12	Female	City
II	Female	58	1.5	Purchase of a tool	12	Female	Rural
III	Male	76	2	Modification of pharmacotherapy for PD (adverse effects)	12	Male	City
IV	Male	67	1.5	Admission to hospital (acute), modification of pharmacotherapy for PD	12[a]	Male[b]	City
V	Male	65	1.5	Involvement of speech therapist	12	Male[b]	City
VI	Male	79	4	Admission to hospital (acute)	12[a]†	Female	City
VII	Female	75	1	Consultation of GP, admission to hospital (planned investigation)	6	Female	Rural
VIII	Male	73	2	Domestic adjustment, modification of pharmacotherapy for PD (adverse effects)	6	Female	City
IX	Female	70	4	Consultation of GP, consultation of neurologist, modification of pharmacotherapy for PD (adverse effect)	6	Male	Rural
X	Female	64	2.5	Modification of pharmacotherapy for PD	6	Male	Rural
XI	Male	65	1.5	Consultation of GP, ENT specialist and neurologist, modification of pharmacotherapy (laxative)	6	Male	City
XII	Male	72	2	Consultation of neurologist, consultation of GP, modification of pharmacotherapy (laxative)	6	Male	Rural

[a] Did not finish follow-up
[b] Same GP
†Deceased during follow-up

Table 3. Characteristics of the participating patients with Parkinson's disease and their general practitioners
- including an oversight of the encountered care transitions -

Role of the GP in PD care

A total of 36 codes concerning the role of the GP in PD care were identified in the analysis. These codes were grouped into 10 different themes: self-reliance and autonomy; diagnosing PD; follow-up prescriptions; acute care; lack of expert knowledge and skills; awareness of experiences and well-being; careful monitoring; patient's personal situation; optimizing care and palliative care. The analysis gave insight into two different roles for the GP, depending on the stage of PD (Additional file 1).

Patients described a few specific roles for the GP in PD care in the early stages of the disease. GPs reiterated these roles and added others. Patients did not express any specific roles in more advanced-stage PD, whereas GPs mentioned additional roles in case of disease progression.

Limited, reactive role in early-stage PD care

Patients preferred and often succeeded to self-manage the care transitions they encountered.

I don't go to the GP for every little thing. If I can solve it myself, then I'll just do it. (Patient XII, male, 72 years, H&Y 2)

GPs underlined the importance of self-reliance and autonomy in decision-making.

I think it's good if people take the initiative; I believe self-reliance is after all very important. So, if people do that, I can only encourage it. (GP of Patient II, female, rural)

As long as someone is of sound mind, and can say what he wants to say, I think it borders on patronizing to try and take over. […] It has to do with instinct and autonomy and things like that. (GP of Patient XII, male, rural)

Patients described a few specific roles for the GP in the early stages of PD. One task both patients and GPs agreed on is recognition of the disease, although they acknowledged that this could be difficult.

First of all, he [the GP] has to be able to diagnose [PD]…and he wasn't able to do that when I went. […] It's really difficult for the GP, because PD isn't a disease where you see patients with all the same symptoms. (Patient XII, male, 72 years, H&Y 2)

The most important role is to recognize it, to diagnose. […] The start of Parkinson's is often very vague and it is important for a GP to recognize it in time. (GP of Patient X, male, rural)

Furthermore, patients stated that they expect their GP to provide follow-up prescriptions for PD medication.

The medicine is finished after three months, so that's when I have a reason and I plan to visit the GP. (Patient I, male, 65 years, H&Y 1)

GPs added a role in pharmacosurveillance to this.

I want to know what medication a patient is taking, so I know the correct dose if I write a repeat prescription […] but also to have a good overview of the possible interactions or side effects. (GP of Patient I, female, city)

In addition, patients indicated that they would turn to their GP in case of an acute care transition.

If something happens, if I fell off this chair, then she [the GP] will probably come. Not the specialist, that one stays where he is. (Patient VIII, Male, 73 years, H&Y 2)

For all other PD-related questions and transitions, patients preferred not to contact the GP, since they felt that the GP lacked expert knowledge and skills. Patients chose to turn to specialized care directly.

If I want my shoes fixed, I will go to the shoemaker. So, now I have Parkinson's, I go to the neurologist. (Patient III, male, 76 years, H&Y 2)

She [the GP] knows of Parkinson's but doesn't know that much about it; she's not an expert so I don't want to trouble her. (Patient I, male, 65 years, H&Y 1)

In line with this, GPs expressed reluctance to be involved in PD care, because they did not feel competent.

I: What's the difference between Parkinson's and other chronic disorders, where you do have a central position as GP?
GP: The prevalence. I think Parkinson's is quite a complex disease with different manifestations […]. It's complicated in terms of pharmacotherapy […]. It's something I don't have much experience with; there are plenty of things I feel more familiar with. So, I figure, if the neurologist can handle it well, then I prefer to leave it to him. (GP of Patient XII, male, rural)

Although GPs described their role in the early stages of PD as limited and reactive, they also mentioned that patients might not realize what accessory role the GP could have.

I suppose he [the patient] is content with the current care, and just doesn't realize [what role the GP could have]. (GP of Patient V, male, city)

GPs illustrated their role by mentioning the importance of being aware of patient's experiences and well-being.

The GP doesn't have a big role in the treatment of PD, but you have to be aware of what is happening […], there's more to the patient than just PD. (GP of Patient IX, male, rural)

Therefore, GPs emphasized that it was important to stay in touch with the patient at any time during the course of the disease.

The GP should stay in touch with PD patients, since the consequences of the 'care' part […], especially for patients with more progressed disease, are considerable. (GP of Patient V, male, city)

Extended, proactive role in advanced-stage PD care

Patients with mild-stage PD did not express specific roles for the GP in case of progression of the disease. In addition, the patient with severe disease and the caregiver of the deceased patient (H&Y 4) did not describe roles for the GP other than the above-mentioned. GPs however mentioned a shift in their role.

In the beginning of Parkinson's, everything can be easily handled by the neurologist and the specialized nurse. But when people develop more incapacitating complaints…you know, a neurologist doesn't do home visits…So I think, the more the disease progresses, the more important our role will become. (GP of Patient VII, female, rural)

GPs mentioned careful monitoring and paying attention to possible care transitions as important tasks. Moreover, they described a proactive role in discussing possible solutions for signaled problems, while taking into account the patient's care preferences.

I notice things that in my opinion could be better. I explain that to them and it is discussed. Then it is up to them to do something with it or not. (GP of Patient III, male, city)

In addition, GPs expressed they would consider possible benefits from a home visit, such as insight into patient's living circumstances.

Sometimes a home visit can be very enlightening, to get insight into the possibilities at home. […] Or if people really want a private conversation out of the medical setting… […]. Then I have the idea that that improves the quality of care, because it reinforces the trust of the relationship. (GP of Patient V, male, city)

Moreover, GPs saw an important task in coordinating health care providers and in fine-tuning care with the neurologist.

If more care is needed, then it is my experience that I get called on more often, […] to instigate things, to coordinate. (GP of Patient V, male, city)

If palliative care became necessary, GPs no longer felt reluctant to be involved in PD care. They felt they were the most important health care provider then.

The palliative part […] we can of course offer that in the home situation as no other can. (GP of Patient XII, male, rural)

Discussion

Summary of the main findings

Patients express a preference for self-management of care transitions and autonomy in decision-making. GPs choose to stimulate this behavior by taking a limited, reactive position in early-stage PD care. Moreover, they feel insufficiently competent to extend their role. Patients also feel GPs lack expert knowledge and skills; they focus on their neurologist for PD care. In addition, GPs observe that patients might not realize that GPs could have an accessory role in being aware of patient's well-being. Although patients do not report additional roles for the GP in advanced-stage disease, GPs describe a more extended role as monitor and coordinator of care. Moreover, they feel more confident in their role in progressed disease, especially when palliative care becomes necessary.

Comparison with existing literature

The patients in our study express a favor for self-management of care transitions. Self-management in chronic disease can be described as the degree to which a patient is able and willing to control daily life through management of symptoms, treatment and consequences of a disease.[16] This definition points out the influence of patient's context and comorbidity on self-management.

If self-management is not possible, patients prefer to turn to health care providers specialized in PD care. The focus on expert health care providers for disease-specific care is in line with other research.[17, 18] However, patients' expectations of their GP differ from holistic care for cancer patients[17] to a role limited to providing follow-up prescriptions and offering prompt access to care for patients with cystic fibrosis (CF).[18] The limited role description of CF patients corresponds with our results.

The GPs in our study also see a limited position for themselves in early-stage PD care. On the one hand, this is a purposely chosen position to stimulate patients' self-reliance and autonomy. On the other hand, it is the result of two factors. GPs suggest that patients might not realize that GPs could have an accessory role. The GPs emphasize the importance of being aware of patient's experiences and well-being, thereby describing the GP's role in care as one focusing on person-centeredness. At the same time, GPs are reluctant to fulfill this role.

They seem to be intimidated by the low prevalence and diversity of manifestations of PD. The pharmacological treatment regimen of PD, which is experienced by GPs as complex up until the moment of palliative care, adds to GPs' reluctance. However, these disease-specific characteristics do not particularly influence person-centered care, yet they accentuate the importance of involvement of health care providers specialized in PD.[7]

Person-centered care provided by the GP could add to PD care as well by offering self-management support, thus contributing to patient's autonomy.[16] GPs can use the regular visits of patients to repeatedly invite them to voice their care preferences, preferences that may change over the course of the disease and as a consequence of changes in personal circumstances. Awareness of the patient context and comorbidity allows the GP to help patients prioritize their preferences and to personalize care.[7] This way patients build confidence and feel empowered to voice their preferences in different care settings as well[19], what is especially important since patients nowadays have a central role in decision-making in specialized PD care as well.[20] Moreover, attention for patient care preferences and personal context facilitates the GP in providing person-centered care in more advanced-stage PD as well.

Therefore, community-dwelling PD patients benefit from shared care in which generalist and specialized care reinforce each other and offer personalized care based on patient preferences.[7]

Strengths and limitations of the study

To the best of our knowledge this is the first study that explores what role community-dwelling PD patients and their GPs see for the GP in PD care. The design of the study has several advantages. The monthly visits of the research assistant enabled us to quickly respond to encountered transitions and to interview patients and GPs shortly after the transition, thereby limiting the risk of recall bias. Moreover, the interview prompts, based on the patient videos, facilitated recall if necessary and contributed to questioning in more detail. All interviews were performed by the same skilled interviewer, who took the information from the patient interview into account when interviewing the GP. The interviewer had no professional relationship with the GPs or patients, was experienced in qualitative research and was also part of the analyzing team. Data collection and analysis continued until saturation was reached. We feel confident that we were able to gain comprehensive and in-depth insight into patient and GP perspectives on the role of the GP in PD care.

However, some limitations need to be considered as well. Despite of our effort to collect longitudinal data, our results do not enable description of changes in time of patients' opinion concerning the role of the GP. The decision to reduce the follow-up period from 1 year to 6 months may have contributed to this. Interviews at predetermined moments would likely have resulted in more interviews per patient. However, it is uncertain if a longer follow-

up period or interviews at fixed moments would have led to different results. Furthermore, despite purposive sampling we included more men and patients with mild-stage disease. Although we did include women and patients with advanced-stage disease, it is possible that the roles these groups see for the GP in PD care are not fully explored. Our population, finally, came from a single region in the Netherlands with well-organized specialized PD care, possibly influencing the role patients see for the GP.

In conclusion

Patients and GPs describe a limited role for the GP in early-stage PD. However, community-dwelling PD patients would benefit from shared care in which generalist and specialized care are valued for their specific qualities of person-centeredness and disease-specific expertise. GPs should realize the added value of their generalist approach to care for patients with a complex chronic disorder as PD and fulfill their role with confidence.

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Additional files

Additional file 1

Oversight of all codes and themes concerning the role of the general practitioner in Parkinson's disease care as expressed by patients and general practitioners.

Stage of disease	Role of the GP	10 themes	36 codes	Expressed by
Early	Limited		(Initially) limited	Pt, GP
	Reactive		Being easily accessible	Pt, GP
			Responding to request for help	Pt
			Considering reactive or proactive role	GP
	Reactive	Self-reliance and autonomy	Considering consulting GP	Pt
			Considering patient's ability to initiate and manage	GP
			Role depends on patient's coping	GP
			Discussing pro's and con's of patient's coping	GP
			Discussing role of the GP with patient	GP
	Reactive	Diagnosing PD	Physical examination for new symptoms	GP
			Recognizing possible PD	Pt, GP
	Reactive	Follow-up prescriptions	Providing follow-up prescriptions	Pt, GP
			Pharmacosurveillance	GP
	Reactive	Acute care	Providing acute care	Pt, GP
			Ensuring continuity of care	Pt, GP
	Limited	Lack of expert knowledge and skills	Not for PD-related pharmacotherapy decisions	Pt, GP
			No expert	Pt
			Feeling of incompetence	GP
			Depends on knowledge and experience	GP
	Limited	Awareness of experiences and well-being	Stay in touch with the patient	Pt, GP
			Being informed	Pt
			Paying attention to experiences and well-being	Pt, GP
			Showing compassion/empathy/support	Pt, GP
Advanced	Proactive		Considering reactive or proactive role	GP
	Proactive	Careful monitoring	Being aware of comorbidity	GP
			Monitoring of PD and possible care transitions	GP
			Discussing solutions to signaled problems	GP
	Extended	Patient's personal situation	Taking into account patient's care preferences	GP
			Considering benefit of home visit	GP
			Paying attention to patient's personal context	GP
			Paying attention to the patient-doctor relationship	GP
	Extended	Optimizing care	In dialogue with the neurologist	Pt, GP
			Knowledge of care possibilities	GP
			Offering help in care decisions	Pt, GP
			Coordinating health care providers	GP
	Extended	Palliative care	Main role in palliative care	GP
			Terminal care	GP

Pt = patient, GP = general practitioner

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8

General discussion

The aim of this thesis was to both examine and contribute to patient-centered quality care for community-dwelling patients with Parkinson's disease (PD), starting from the first recognizable symptom and extending into advanced-stage disease. For this, we needed to gain insight into patient experiences before, during and after the diagnosis, as well as the challenges that general practitioners (GPs) are confronted with in providing care to these patients. In this final chapter, we will first give a brief summary of the main results, before discussing these results in light of patient-centered primary care. Subsequently, the chosen methods will be reviewed critically and future perspectives will be discussed.

Overview of main findings

Towards the diagnosis of Parkinson's disease

The qualitative analysis of a purposive sample of patient essays about the pathway from the first recognizable symptom to the diagnosis of PD presented in ***Chapter 2***, clarifies that patients experience difficulties recognizing the symptoms as their knowledge of PD is usually limited to the image of the disease that is presented in the media. This image is generally based on public figures with advanced-stage PD. Because the symptoms that patients experience in early-stage disease are often limited and not very disease-specific, patients tend to find other explanations for them and frequently do not seek help. A patient's decision to seek professional help if symptoms do not resolve or if they progress over time is influenced by a variety of factors such as the involvement of significant others, the patient's previous positive or negative experiences with health care providers and the patient's fear that the GP might think he/she is dramatizing. After the consultation with the GP, patients describe different referral experiences, varying from immediate referral to the neurologist to consultation of several health care providers.

The quantitative analysis of all patient essays, described in ***Chapter 3***, reveals the possible consequences of the difficulties GPs may have referring PD patients, due to the often non-disease-specific prodromal symptoms: one in every seven PD patients explicitly mentions they are dissatisfied with the experienced diagnostic pathway. Patients who feel that their doctor caused delay run a greater risk of being dissatisfied than patients who feel their pathway was not delayed. In addition, the quantitative analysis shows that patients' dissatisfaction with the diagnostic pathway is related to a low level of education, to one patient's request for a second opinion and to female sex.

We used data from the Continuous Morbidity Registration (CMR) database to compare the experienced symptoms and medical consumption of people who were later diagnosed with PD with that of healthy controls. In ***Chapter 4*** we present the results of this study in which we found that PD patients consult the GP more often in the 2 years prior to the diagnosis with symptoms such as constipation, hyperhidrosis and sleep disorders and that over half of these patients present with more than one prodromal symptom in that period. In addition, these

patients are more often diagnosed with Medically Unexplained Physical Symptoms (MUPS) than the controls. Patients also tend to be referred more than usual in the period leading up to their diagnosis.

Changes in care after the diagnosis of PD

In ***Chapter 5***, we describe the design of the longitudinal study on the changes in care encountered by community-dwelling PD patients. We followed patients for 6 months to 1 year and asked them to make video's describing their current physical and mental state. The content of these videos was used to customize the topic guide for the interviews that took place after every encountered care change. In these interviews, we not only asked the patients what changes they experienced and how they coped with them, but also asked them to describe what they expected of their GP. Subsequent to the patient interviews, we interviewed the GPs, to clarify the role they saw for themselves in PD care.

Patients' experiences and coping strategies with changes in care are presented in ***Chapter 6***. The patients prefer to solve the problems they encounter themselves. Patients' self-management is facilitated by their ability to anticipate changes, which is easier if patients have sufficient knowledge of the course of the disease and the related care changes. Being able to self-manage a change gives them a sense of control. Moreover, self-management is related to realistic expectations and acceptance of the post-change situation, even if impairments are not remedied.

Chapter 7 illustrates the roles patients and GPs see for the GP in PD care. Patients expect their GP to recognize PD and refer correctly. Moreover, they rely on their GP in case of an acute care need, and turn to their GP for follow-up prescriptions for PD medication. For all other PD-related questions and changes in care, patients turn to specialized caregivers directly as they feel that GPs lack expert knowledge and skills. The GPs agree on assuming a limited role for themselves in early-stage disease. On the one hand, they purposely choose this position to stimulate patients' self-reliance and autonomy. On the other hand, their limited role is the result of GPs' reluctance to be involved more. The GPs do mention that, despite their limited role, it is important to stay in touch with PD patients at any time during the course of the disease and to take patients' preferences into account because of the considerable impact of the disease on patients' lives. The GPs describe a more extended, proactive role in advanced-stage disease, consisting of careful monitoring, signaling problems such as upcoming changes in care, discussing possible solutions and coordinating health care providers. Finally, the GPs mention they are the most important health care providers when it comes to palliative care.

Patient-centered primary care for PD: reflections on the findings

Based on the results described above, we identified four related themes with important implications for care in all stages of Parkinson's disease: the stereotyped image of PD; the diagnostic uncertainty related to PD; the added value of generalism to PD care; and the

importance of a trusting patient-doctor relationship. These themes will be discussed in greater detail below.

The stereotyped image of PD

As a consequence of the limited representation of PD, such as described by the patients in our essay study, lay persons and health care providers may have a stereotyped image of the disease. An earlier study showed that the general population mainly relates PD to tremor and stiffness, and that there is a widespread belief that the disease presents in older male patients, progresses rapidly and can only be partially controlled by treatment.[1] Moreover, lay people considered their own chances of developing PD as minimal.[1, 2] It is understandable that such a stereotyped image can have a negative effect on help-seeking behavior.[2] A retrospective study amongst PD patients showed that the time from symptom onset to the diagnosis of PD was longer for women than for men.[3] The authors mentioned many factors that might have contributed to this difference, including a possible difference in the experience of prodromal symptoms and the possibility that physicians have a predisposed perception that PD is more likely to occur in men.[3] As we focused on patient dissatisfaction, we do not know if there are differences in experienced delay between the men and women in our essay study, yet we did find that women are more likely to be dissatisfied with their diagnostic pathway. It is possible that underlying this dissatisfaction is the initial response of family, friends and health care providers to normalize female patients' complaints, giving them the feeling they are not being taken seriously. In addition, the belief that the diagnosis of PD will only be made from a certain age onwards may cause difficulty in younger patients to accept this diagnosis, the more so as they will be facing people's prejudice about their inabilities although they might be able to function at near comparable level for years if they are treated adequately.[1] Fortunately, famous young PD patients such as the American actor Michael J. Fox and the English football player Ray Kennedy are helping to break this stigma.[4]

PD can limit patients' abilities to communicate non-verbally. With progression of the disease, patients' volume of speech may become weaker, and facial masking is likely to appear.[5, 6] Patients' limited ability for non-verbal communication may be incorrectly interpreted as antisocial, cold, uninterested or incompetent.[5-7] The impact of facial masking on intimate relationships, however, differs between men and women.[7] This is understandable when taking into account the existing gender role beliefs that are rather similar across cultures.[8] Women are believed to be more emotionally expressive and socially oriented than men.[7, 8] Therefore, female PD patients' inability to express social involvement non-verbally due to facial masking is more inconsistent with people's expectations and will be interpreted more negatively than the same inability would in males.[7, 8] Although knowledge and understanding of the disease is greater in GPs and other health care providers, research has shown that they may also have difficulty realizing that their initial assessment of a PD patient's abilities, based on their first impression of the patient, may not be reliable.[5, 6] Knowing that a significant part of newly diagnosed PD patients already manifest cognitive dysfunction and that PD

patients without dementia may already have trouble with medical decision-making, a first non-verbal impression of incompetence can, without further consideration, contribute to the underestimation of PD patient's ability to self-manage. However, our study shows that community-dwelling PD patients in early stages of their disease are able to self-manage changes in care. Health care providers, therefore, should be aware of the risk that their assessment of PD patients' abilities may be prejudiced and take this into account.[6]

The diagnostic uncertainty related to PD

The caseload in a general practice is usually undifferentiated and unorganized.[9] Every patient has a small but real chance of suffering from a serious condition, and patients generally present diseases in non-textbook ways.[10, 11] For GPs it is a challenge to deal with the diagnostic uncertainty that is part of their daily work.[9, 10] For patients, the uncertainty involved in awaiting diagnosis can have a tremendous impact on their daily life, as we know from a study of dementia patients and their caregivers.[12]

Wray describes two types of diagnostic uncertainty: informational uncertainty and intrinsic uncertainty.[13] Informational uncertainty is the type of uncertainty that is related to shortcomings in disease-specific knowledge and skills and to a flawed patient-doctor relationship, lacking insight into patients' care preferences.[13] The results presented in this thesis already described that recognizing a pattern of symptoms pointing to PD may be difficult for GPs because they have limited knowledge of the prodromal and early-stage symptoms of the disease. The importance of a trusting patient-doctor relationship will be discussed below.

Intrinsic uncertainty is the uncertainty that exists because the clinical course of a disease cannot be predicted, as diseases may present and progress in markedly different ways in different patients.[13] The initial presentation and the clinical course of PD are highly variable: some patients with classic PD symptoms are diagnosed straightaway, whereas others experience a long and difficult diagnostic pathway. Although uncertainty about the PD diagnosis cannot be taken away completely until after death – after all, the diagnosis can only be fully ascertained post-mortem – the GPs' response can make a difference. GPs may turn to overdiagnosing presented symptoms for fear of missing a serious condition [14], and if they do so for only one or two of the symptoms instead of creating an overview, this could lead to tunnel vision in the wrong direction.[11] The difference we found in the MUPS diagnoses between the patients and the controls in our case-control study may, in fact, not reflect a difference in MUPS prevalence but may reflect GPs' diagnostic uncertainty with symptoms that are difficult to grasp, such as symptoms of autonomic dysfunction. Patients with cancer and Amyotrophic Lateral Sclerosis describe over-reassurance as another response of GPs: 'Don't worry; there is nothing wrong with you'.[15-17] As a result, they get false hope whenever an examination excludes a worrisome diagnosis and tend to normalize new symptoms or attribute them to an initially benign diagnosis.[15-17] Cancer patients also mention the possibility

of under-support and its consequences: patients who initially had the experience they were sent away or were not being taken seriously, feel uncertain if and when to present their symptoms again since they do not want to appear to be hypochondriacs.[16] Over-reassurance and under-support are also described in our study, causing the risk of diagnostic delay.

There is extensive research on strategies to deal with diagnostic uncertainty in a way that prevents delay as much as possible.[9, 10, 13, 16, 18] In all strategies, patient-doctor communication is essential. Diagnostic safety netting is one of these strategies.[10, 16, 18] It refers to an approach of openly sharing the uncertainty of the diagnosis with the patient and indicating that more visits to the GP might be necessary before the signs and symptoms of the condition become more evident.[16, 18] Multiple studies show that patients value health care providers who recognize the limits in their own knowledge and skills and act upon it.[17, 19] However, the way in which diagnostic uncertainty is shared needs to be carefully considered. An explicit verbal expression, such as 'I don't know', is known to undermine patients' confidence although physicians generally underestimate this effect.[9, 13, 20] In addition, adding some reassurance might be helpful to reduce patients' stress while facing an uncertain situation.[13] An important feature of diagnostic safety netting is the instruction to patients to return to their GP whenever they are worried or, if possible, when symptoms develop in a certain direction.[10, 16, 18] This way, patients are empowered to reconsult, and the experience of under-support can be minimized.[18] The fact that the lower educated patients in our essay study are more likely to be dissatisfied with their diagnostic pathway than the higher educated patients, however, might reflect the difficulties low-educated patients have with understanding their GPs' explanation and acting upon it. In the case of a complex clinical disease course, therefore, GPs should take the patients' level of health literacy into account in either aiming to fully explain the course of the disease or mainly to focus on sharing the diagnostic uncertainty. In addition, it is essential for GPs to keep an open mind on any possible diagnosis in order to recognize if patients present new symptoms or if symptoms develop in a way that does not suit the first diagnostic idea.[11]

A strategy that can be used in combination with diagnostic safety netting, especially in the case of a slowly developing disease without life-threatening symptoms such as PD, is watchful waiting.[9, 10, 16] GPs are in the position to use this approach as patients will generally first present their symptoms in a consultation in general practice. The symptoms presented in subsequent visits can then be combined into one image that is clear enough to consider referral, as in the case of Mrs P.D. described in the preface. As it is known that cancer patients feel uncertain and anxious if referral is not sufficiently explained to them and that PD patients' dissatisfaction with the way the diagnosis is expressed affects their experienced quality of life, it is clear that referral should be discussed with patients, with an explanation of the considerations that is tailored to patients' level of health literacy.[9, 21-23] A study among patients with MUPS shows that watchful waiting, if applied in a proper way, does not have to lead to patient dissatisfaction.[24] The same strategy might also prevent patient dissatisfaction with the diagnostic pathway of PD.

The added value of generalism to PD care

It seems fair at first that the patients in our longitudinal study focus on the movement disorder specialist. After all, the medical specialist has the specific knowledge and skills that are essential for treating a complex disease such as PD, with a diverse expression and a complicated treatment regimen.[25-28] Patients with cancer and Multiple Sclerosis also feel that their disease should be treated by a medical specialist [29, 30], but they also mention another role for the GP that is at least as important and is an essential component of chronic disease care: the GP should take care of the patient as a whole person.[29-31] Although the patients in our study do not describe this role, the GPs do: 'there's more to the patient than just PD'. GPs may not be experts in PD-specific care, but they are experts in whole person medicine.[32]

The expertise of generalism has three clear characteristics: a continuous rather than an episodic view; the integration of biomedical knowledge and knowledge of the patient's unique situation; and the possibility to support decisions that recognize health as a resource for living rather than as an end in itself.[25, 32] A GP's continuous view, the first characteristic of generalism, offers the advantage of the watchful waiting strategy, described above, during the diagnostic pathway of PD. Moreover, it allows for continuity of care. Freeman describes two types of continuity of care.[33] One is relationship continuity or personal continuity, which is most valued in general practice and helps patients to build trust in their GP.[19, 34] The other type of continuity is management continuity, for which communication of relevant information and cooperation between the health care providers involved are essential.[33, 34] Patients with PD do not experience their care needs in episodes of care or in different care settings.[33] For them, the changes in care and related health care needs that come with progression of the disease are continuous.[35] Management continuity, therefore, is indispensable. Patients generally prefer one health care provider to coordinate care, and earlier research in the Netherlands showed that chronically ill patients appreciate it if their GP fulfils this role.[36] The patients in our study do not explicitly describe such a role for the GP, but the GPs underline their role in management continuity and also describe a role in care planning.

The added value of the GP as the coordinator of the health care providers involved in PD care lies in the second characteristic of generalism: the integration of knowledge of a patient's disease and his/her unique situation.[33] The position of Dutch GPs as family doctors gives them insight into the past and current physical and mental state of patients and family members and into the contextual circumstances that influence their well-being.[37] GPs also have the possibility of making home visits, which is especially valuable for PD patients as there is a known discrepancy in performance of patients in the complex living situation at home and in the well-lit and wide corridors of a hospital.[38] In case of the need for coordination of care, GPs can use their insight into the patient as a whole person to integrate general care and disease-specific care into personal patient-centered care that prioritizes problems.[26, 39, 40] Their long-term knowledge of patients also facilitates their assessment of patients' ability to adapt, which is important, as this ability to adapt and to self-manage determines their

experienced health.[41] Moreover, the ability to adapt is assumed to influence the need for health care interventions more than the disease itself.[42] The results of our study are in line with this.

The GPs' holistic view also allows them to support their patients in formulating self-management goals that are not disease-oriented but focus on what is most meaningful to patients, the final characteristic of generalism.[25, 43-45] Concentrating on the most pressing issues rather than the disease itself is relevant, as PD patients are known to differ in their perception of the most troublesome problems that come with the disease.[39, 46, 47] In addition, patients are likely to have comorbidity.[48] Self-management needs and goals of different diseases may be conflicting, necessitating a focus that goes beyond the single disease level.[45, 48-50] As understanding the patients' needs for support may improve its effectiveness, frequent review of these needs is important.[51] GPs have the possibility to use the consultations of their PD patients, regardless of the reason of consultation, to try to understand the meaning PD has in the patients' lives and to invite them repeatedly to voice their needs and preferences.[50] They are able to tailor their support to a patient's unique situation, therefore, based on the disease stage, comorbidity, personal context and care preferences that are relevant at that time.[45, 50] The advantage of this approach goes beyond primary care. If patients are supported to formulate their self-management goals in a familiar setting with support offered by a GP who knows most of the relevant aspects, they may also feel empowered to do so in different care settings.[44] This is especially important for PD patients because they now play a central role in decision-making in specialized PD care as well, although a recent study showed that patients experience difficulty mentioning their needs.[52, 53] If patients are able to voice what problems are most pressing to them and what goals they value most, the therapeutic decisions made are likely to correspond with their preferences.[39]

The importance of a trusting patient-doctor relationship

As described above, relational continuity is one of the characteristics of primary care. Relational continuity – the ongoing therapeutic relationship between patients and their GPs – is essential to effectuate the other characteristics described above.[34] For such a relationship, physicians need to possess good communication skills and be caring.[54] Showing empathy, in addition, contributes to openness in the relationship and to feelings of safety and trust.[55] The inherent knowledge and power imbalance that exists between patients and doctors underlines the importance of trust.[56, 57]

PD patients will benefit from a trusting relationship with their GP from the moment they become aware of the first prodromal symptom. In our essay study, we describe the effect previous negative experiences in communication with health care providers may have on patients' help-seeking behavior. In addition, empathic communication and trust in the GP leads to greater patient willingness to open up and express symptoms, expectations and emotions such as uncertainty, worry and frustration.[31, 58, 59] PD patients should also trust that GPs will

try to understand the first symptoms, which may sometimes be confusing or misleading.[9] One patient in the essay study describes he always felt taken seriously by his GP, even though the symptoms he presented were vague. Moreover, a trusting patient-doctor relationship can empower PD patients to deal with the uncertainty that is inherent in their diagnostic pathway, for example, when patients feel that the GP takes the diagnostic journey together with them.[9] From a study in cancer patients, it is also known that a trusting relationship may reduce fear of medical error and leads to more patient satisfaction.[56] A trusting relationship between PD patients and their GPs can also help patients to deal with the unpredictability and uncontrollability associated with progression of their disease.[60-62] Although the patients in our longitudinal study do not mention that they expect the GP to keep an eye on them, the GPs do express this as being one of their roles. GPs in a study of patients with multimorbidity mentioned that a trusting patient-doctor relationship facilitates the management of complex health care situations.[50] Patients with multiple chronic conditions wanted their GP to stay in touch with them and to offer guidance during progression of the disease(s).[36, 62] The same is true for cancer patients, who interpret absence of regular contact as a lack of interest of the GP.[63]

In line with earlier research, the patients and GPs in our study describe the importance of autonomy and self-management, leading to a sense of control.[64] The preference for self-efficacy in the early stages of PD could suggest that a trusting relationship between patients and GPs is not essential. The opposite, however, is true. A trusting relationship facilitates patients' self-management.[55, 58] The support GPs can offer in self-management has already been outlined above. PD patients may not be fully aware, however, of the added value of the GP in this respect, as one GP described in the interview study. With further progression of the disease, PD patients will increasingly experience difficulties with medical decision-making as their ability to organize information and to understand treatment options becomes more and more impaired.[65] Patient autonomy can then be maintained by stimulating shared decision-making.[58] The central role of a trusting relationship with the GP in this, and, more explicitly, the importance of GPs' knowledge of patients' personal circumstances, has already been described by PD patients in an earlier study.[54] Patients in general, however, are inclined to take a passive decision-making role out of fear they will annoy health care providers and receive lower quality care.[57] Although the role of the GP may shift, therefore, from self-management support to continuous counseling in stimulating shared decision-making, a trusting patient-doctor relationship remains equally important to encourage patients to voice their preferences.[31, 39, 54, 56]

Methodological considerations

The qualitative methods

We used qualitative research methods to gain insight into patients' experiences with the diagnostic pathway of PD, to gain insight into the experiences and coping of community-dwelling PD patients with changes in care encountered in the course of PD, and to clarify the

role these patients and their GPs see for GPs in PD care. In this section, we will discuss the strengths and limitations that are specific to the qualitative analysis of essays (***Chapter 2***) and the application of qualitative interviews, including the use of video diaries (***Chapter 6 and 7***).

The format guiding patients in retrospectively describing their experiences of the diagnostic pathway of PD led to essays that were similarly structured (***Chapter 2***). The open questions used in this format, on the other hand, allowed patients to decide freely on the content and the amount of detail they wanted to provide. We believe that the content of the essays, therefore, reflects what matters most to the patients. Using an online approach allowed us to reach a large target group, as opposed to face-to-face interviews. As a consequence of this decision, patients who feel uncomfortable or are unable to use the computer are left out. Nevertheless, we tried to get a broad and reliable view of patients' experiences by using, first, an inductive approach to our analysis of the essays of a highly variable sample of respondents, and, subsequently, a deductive approach to the analysis of essay contents of a second, purposive sample of respondents.[66]

The longitudinal character of the study of community-dwelling PD patients and their GPs and the monthly visits of the research assistant enabled us to interview patients and their GPs at turning points in life, namely shortly after the patients encountered a change in care, rather than at fixed moments (***Chapter 6 and 7***).[67] The risk of recall bias, therefore, is limited. In addition, we used information from the patient videos as interview prompts, thus allowing us to question patients in more detail. All interviews were performed by the same skilled interviewer, who could take the information from the patient interviews into account when interviewing GPs. Nevertheless, although we selected a purposive sample of patients to participate in our study, relatively few women and patients with advanced-stage disease were included. For practical reasons, the patients and GPs came from one single region, in which specialized PD care is well developed. In addition, despite the longitudinal set-up, only few patients encountered more than one moment of change in care during the study period. Therefore, our data do not allow us to describe changes in time of patients' experiences, coping or opinions on the role of the GP in PD care.[67] It cannot be excluded that a different study sample, another study region or a longer follow-up period would have led to different results.

The quantitative methods

A quantitative analysis approach was chosen to gain insight into factors influencing patient dissatisfaction with the diagnostic pathway of PD (***Chapter 3***) and to characterize the prodromal symptoms of PD presented in general practice (***Chapter 4***). Here we will describe the specific strengths and limitations of the methods used. The quantitative data that were collected as described in ***Chapter 5***, are not included in this thesis. The collection methods of these data will therefore not be reviewed here.

We used an original approach to mixed methods research to enable content analysis of more than 900 freely written patient essays.[68] The strengths of the data gathering method have been described above (***Chapter 2***), and the coding format used for the study presented in ***Chapter 3*** was based on the results of the qualitative analysis. As the inter-observer agreement on the independent coding of patient dissatisfaction by two researchers in a random sample of 225 of the essays can be considered almost perfect, we are confident that we used a reliable data extraction method.[69] However, interpreting spontaneous reports limits the interpretation of causality. In addition, factors that might be relevant but have not been described – such as earlier negative health care experiences – will remain unknown.

The most important strength of the nested case-control study we performed on the prodromal symptoms of PD (***Chapter 4***) is the use of the CMR database.[70] This well-documented database allowed us to include unselected cases of PD and match them to controls from the general population. In addition, information from the database is not biased by a retrospective view. Nevertheless, the low incidence of PD limits the number of patients that could be included, and the lack of presentation of prodromal symptoms by patients or reporting by GPs may have led to an underestimation of the actual prevalence of the prodromal symptoms studied. However, focussing on the symptoms patients present has improved our understanding of the symptoms that patients find significant enough for help-seeking and that GPs encounter in their everyday clinical practice.

Future perspectives

Recommendations for clinical practice

Using a watchful waiting strategy with safety netting during the diagnostic pathway of PD has the potential of limiting diagnostic delay and, in particular, patient dissatisfaction. Because not all presented prodromal symptoms will immediately lead to a diagnosis of PD, and because patients with other conditions might benefit from the suggested approaches to diagnostic uncertainty as well, GPs should try to apply these methods in every diagnostic pathway where there is no acute threat to life.

It appears valuable for PD patients to feel they make the diagnostic journey of PD together with their GP, as this can build a strong foundation for the trusting patient-doctor relationship that is much needed with progression of the disease. Although the moment PD is diagnosed is also the starting-point of a new therapeutic relationship between patients and medical specialists, GPs should be aware and feel confident of the continuing added value of their relationship with patients and act upon it. After all, community-dwelling PD patients benefit from shared care, in which generalist and specialized care professionals reinforce each other's expertise and offer personalized care based on patient preferences.

Recommendations for education

Health care providers' and lay people's perception of PD should be modified from the

stereotyped image of an elderly male showing advanced-stage disease symptoms to a more complete representation of the disease, including a realistic view of the prodromal situation and awareness of the potentially incorrect assessment of PD patients' competence. In order to facilitate earlier recognition of the symptom pattern of PD by health care providers, GPs, trainees and other health care providers potentially involved in care during the prodromal stage of the disease – such as physical therapists or orthopedic surgeons – should receive further education on the prodromal presentation of PD. It is important to note, in this regard, that the medical curriculum has so far paid little attention to prodromal PD, as this is a rather novel concept.[71] Moreover, GPs and trainees should be taught about the potential changes in care encountered by their PD patients, in order to be able to guide them.

Knowledge of PD and its early stages should also – and perhaps particularly – be improved in the general population. Not only will this help patients to recognize the seriousness of their symptoms, but it may also help family and friends to encourage patients' help-seeking behavior. To improve public knowledge of PD in the prodromal stage, information campaigns – for example by the government, by professional organizations involved in PD care or by the Parkinson's Disease Association - should be considered. Given the impact television potentially has on image-making, the media can also play an educational role by showing a more representative picture of the disease.

In addition, PD patients should be more effectively educated about the course of their disease and the changes in care they are likely to encounter, as this will help them to think ahead and to have more realistic expectations of the future. The information already existing on www.thuisarts.nl should be enhanced, therefore, to include information on the potential care changes and the role GPs can play in offering support. In addition, patients should be taught to voice what is most meaningful to them and what support needs they have. This way, self-management support is more likely to fit their needs, and care decisions made in shared responsibility between patients and health care providers are more likely to align with patients' preferences.

Recommendations for research

The difficulties PD patients can experience in (non-verbal) communication and medical decision-making may prevent them from expressing their needs for self-management support and may affect their ability to participate in shared decision-making. Merely inviting patients to share their needs and preferences may not be enough to overcome these specific obstacles. Research should be performed into the difficulties PD patients experience in voicing what is important to them, and into how these patients feel they could be trained. Moreover, specific attention should be paid to gender differences in that regard.

Although we were able to gain in-depth insight into community-dwelling PD patients' experiences and coping with changes in care in the early stages of their disease, the results may not represent the experiences and coping strategies of patients with advanced stages

of PD. Such patients may also have different views on the role of their GP. As the general population is ageing and government decisions on health care force patients to live at home for longer periods of time, it is to be expected that GPs will increasingly be facing changes in care of patients with late-stage PD. Insight into their support needs would be valuable to be able to provide tailored care to these patients. Moreover, patients living in different regions with less developed specialized PD care facilities may well have different expectations of their GPs, and the GPs in such a region may differ in their feeling competent to provide care to PD patients. Performing a similar study in a different region with a larger study population including patients with more advanced-stage disease, therefore, would be advisable.

For Dutch GPs, there is a Parkinson's Disease Guideline that can support them in providing care to PD patients.[72] However, none of the GPs we interviewed mentioned this guideline, underscoring the well-known challenge of guideline implementation into clinical practice. As the number of PD patients per general practice is likely to increase in the near future, research should be performed on GPs' awareness of this guideline and on the best way of implementing it in their daily clinical practice. Moreover, the factors facilitating and hindering the use of this guideline may help us understand how GPs can be supported in offering their PD patients the care they need.

Conclusion

The challenging and sometimes lengthy diagnostic pathway of PD leads to uncertainty for both patients and GPs. During the time it takes for the signs and symptoms of PD to become more evident, diagnostic uncertainty should be shared with patients. Using a watchful waiting strategy will allow GPs to recognize a pattern in the symptoms that are presented over time and to suspect PD as soon as possible, while taking care to avoid tunnel vision towards the first diagnostic idea and making sure that patients are offered continuous support and are encouraged to reconsult with new complaints or worries. This way, patients and GPs experience the diagnostic pathway together, which may strengthen the trusting relationship between them.

After the diagnosis has been made, the GPs' role will shift, but the importance of a trusting relationship and the GPs' involvement in PD care do not diminish. Patients prefer to remain autonomous for as long as possible, and self-management of changes in care appears to be possible in early-stage disease. However, GPs continue to play an important role by offering support to patients in formulating self-management goals that are not disease-oriented but focus on what is most meaningful to patients. Knowing the patient as a whole person facilitates this. This knowledge and GPs' possibility to repeatedly invite patients to express what problems are most pressing and what goals are valued most, moreover, allow them to tailor their self-management support to the needs that constantly change with progression of the disease.

The PD patients' ability to participate in medical decision-making will decrease with further disease progression. In order to maintain autonomy for as long as possible, the GPs' role will then shift towards a counseling role in shared decision-making. A trusting patient-doctor relationship remains equally important during this stage, to encourage patients to voice their preferences. The GPs' knowledge of their patients' disease and unique situation allows them to coordinate all health care providers that are involved in care, and to integrate care in a patient-centered manner by prioritizing problems in line with patients' preferences.

It is clear, therefore, that Parkinson's disease in primary care should be a joint journey of community-dwelling patients and their GPs, in which a trusting relationship is of vital importance to offer much-needed personalized support.

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9

Summary
Samenvatting

Summary

Chapter 1 General introduction

In Chapter 1 the rationale, aims and outline of this thesis are described.

Years before patients are being diagnosed with Parkinson's disease (PD), they may already be suffering from –so called prodromal- symptoms. These symptoms are usually not disease-specific. It may be difficult for patients to recognize that the symptoms need medical attention and for general practitioners (GPs) that PD is the common cause of the presented symptoms. Consequently, the diagnostic pathway of PD can be lengthy and uncertain and may negatively affect the patient-doctor relationship. Progression of PD will, in addition, inevitably lead to changes in care that patients have to cope with. Although these changes might be experienced without the involvement of professional health care providers, patients may still benefit from support. GPs are experienced in offering such support to patients with prevalent chronic conditions.

PD patients may benefit from early intervention and treatment. A trusting patient-doctor relationship is indispensable to offer personalized support during changes in care and to enable shared decision-making. Knowledge of the factors that influence the course of the diagnostic pathway of PD and the experiences of patients during this pathway is however scarce. Moreover, most studies on the prodromal symptoms of PD are hospital-based, while GPs are consulted earlier, when symptoms are usually still limited. Furthermore, in order to provide personalized support, our knowledge of PD patients' experiences with changes in care and their expectations of the GP need to be increased. This thesis aims to provide more insight into patients' experiences before, during and after the diagnosis of PD, as well as the challenges GPs are confronted with in providing care to these community-dwelling PD patients.

Towards the diagnosis of Parkinson's disease

The patient's perspective

Chapter 2 Time intervals in diagnosing Parkinson's disease: the patients' views

In order to gain more insight into patients' experiences of the pathway from the first recognizable symptom to the diagnosis of PD, we performed a qualitative study (Chapter 2) of essays about the diagnostic pathway, written by 902 patient members of the Dutch Parkinson's Disease Association. A purposive sample of 52 essays was analyzed independently by two researchers in an iterative process. Saturation in data analysis was reached. We found that the diagnostic pathway of PD is an iterative process that can be divided into three time intervals: recognizing the symptoms; deciding to seek help; and the process of diagnosing PD. Patients described that recognizing the prodromal symptoms of PD can be challenging as their knowledge of the disease is generally based on public figures with advanced-stage PD. The decision to seek professional help if symptoms do not resolve or progress over time is influenced by a variety of factors including the involvement of significant others and the patient's attitude toward health care. Patients also described they had the feeling that

GPs experience difficulties recognizing PD. We conclude that effort is required to increase recognition of the prodromal symptoms of PD by lay people and health care providers in order to facilitate an earlier diagnosis and timely therapeutic intervention.

Chapter 3 The diagnostic pathway of Parkinson's disease: a cross-sectional survey study of factors influencing patient dissatisfaction

In Chapter 3 we present the results of the analysis of all 902 patient essays mentioned in Chapter 2. Based on the qualitative analysis, a coding format was developed to examine the content of the essays. Two researchers independently coded more than 150 essays, initially to create consensus on the coding method, and later to discuss doubts in coding. The other essays were coded by one researcher. The focus of the study was the relationship between dissatisfaction and demographic characteristics, duration of the diagnostic pathway, communication with the GP and neurologist, the number of involved health care providers, whether or not a second opinion had taken place and whether diagnostic delay was experienced by the patient. A subgroup analysis was performed to gain insight into sex-related differences. Of all patients, 16.4% explicitly described they were dissatisfied with the diagnostic pathway, whereas 4.8% were very satisfied. The chance of dissatisfaction increased with a lower level of education, the involvement of more than one health care provider other than the GP or neurologist, a second opinion initiated by the patient and delay caused by a health care provider. When only the GP and the neurologist were involved, women were more likely to be dissatisfied than men. We conclude that GPs can positively influence patients' experiences and contribute to a trusting patient-doctor relationship if they are aware of these risk factors for patient dissatisfaction and pay extra attention to communication and shared decision-making.

The general practitioner's perspective

Chapter 4 Prodromal symptoms and early detection of Parkinson's disease in general practice: a nested case-control study

The nested case-control study presented in Chapter 4 was performed in order to characterize the prodromal symptoms of PD presented in general practice. For this study we used data from the Continuous Morbidity Registration (CMR) database. This database contains information of all health problems that a population of approximately 12 000 patients from four general practices presented to the GP. We included 86 patients diagnosed with PD and 78 matched controls and found that in the 2-year period prior to the diagnosis, PD patients more often presented with functional somatic symptoms, constipation, hyperhidrosis and sleep disorders than controls. Patients also more frequently experienced more than one prodromal symptom and were more often referred within the primary care team or to a medical specialist. We conclude that GPs should be alert when patients present with multiple symptoms in a 2-year period, especially considering the possible benefits of early intervention.

Changes in care after the diagnosis of Parkinson's disease

Chapter 5 Transitions in Parkinson's disease in primary care: protocol of a longitudinal mixed methods study

Chapter 5 contains the protocol of a longitudinal study on changes in care encountered by community-dwelling PD patients. It describes recruitment, data collection and data analysis in detail. A purposive sample of GPs in and around Nijmegen, the Netherlands, and their community-dwelling PD patients without cognitive dysfunction were planned to participate in the study for a period of 1 year. During that year, patients would make a video diary once a fortnight to give a verbal and non-verbal impression of their well-being. Every month, a research assistant would collect these videos and ask the patients if they had encountered any change in care. If so, patients would be questioned on their experiences and coping with the change and on the role they saw for the GP in PD care, using information from the videos to customize the interview topic guide. Patients' GPs would also be interviewed in-depth, focusing on their role in PD care during the encountered change in care and in general. The anonymous, verbatim transcripts of the interviews would be analyzed according to the principles of constant comparative analysis. Inclusion of GPs and patients, data collection and data analysis was planned to continue until saturation in data analysis was reached.

The patient's perspective

Chapter 6 Being in control of Parkinson's disease: a qualitative study of patient experiences

Chapter 6 describes the experiences and coping with changes in care of community-dwelling PD patients, based on the interviews with a purposive sample of 16 community-dwelling PD patients, who participated in the study described in Chapter 5. We found that patients experienced a variety of changes in care such as changes in the level of unpaid care, the purchase of tools, modification of pharmacotherapy or admission to hospital. Three themes had a great influence on patients' experiences and their acceptance of impairments afterwards: anticipating change; self-managing change; and managing expectations. Unforeseen changes in care could be overwhelming for patients, while being able to anticipate change enhanced patients' self-management and feeling of control. Patients preferred to solve the problems they encountered themselves, and if they succeeded in doing so, they had realistic expectations and could accept impairments that remained despite the change. If a change was initiated by a health care provider however, expectations were often unrealistically high and unmet. We conclude that the changes in care PD patients experience are closely related to their personal context. Even when disease-specific care is offered by specialized health care providers, self-management support and help with establishing realistic self-management goals can therefore be offered by GPs, who are well acquainted with the facilitating and complicating factors in their patients' situation.

The general practitioner's perspective

Chapter 7 Parkinson's disease: patient and general practitioner perspectives on the role of primary care

In Chapter 7 we clarify the role PD patients and their GPs see for the GP in PD care. In order to do this, the interviews of the earlier described 16 community-dwelling PD patients and 12 of their GPs were independently coded by two researchers and analyzed according to the principles of constant comparative analysis. We found that patients preferred self-management of changes in care and autonomy in decision-making. GPs chose to stimulate this behavior by taking a limited, reactive position in early-stage PD care. Moreover, they felt insufficiently competent to extend their role. Patients also felt that GPs lack expert knowledge and skills; they focus on their neurologist for PD care. In addition, GPs observed patients might not realize what accessory role the GP could have, a role GPs described as essential in being aware of patient's well-being. Although patients did not report additional roles for the GP in advanced-stage disease, GPs described a shift towards a more proactive and extended role. We conclude that, although patients and GPs see a limited role for the GP in early-stage PD care, GPs should feel more confident of the added value of their generalist approach to care for patients with a complex chronic disorder such as PD. If generalist and specialized care reinforce each other, PD patients benefit.

Chapter 8 General discussion

In Chapter 8 we discuss the results of the performed studies in light of patient-centered primary care. Moreover, we review the methods used and discuss recommendations for clinical practice, education and research.

We identified four related themes with important implications for care in all stages of PD: the stereotyped image of PD; the diagnostic uncertainty related to PD; the added value of generalism to PD care; and the importance of a trusting patient-doctor relationship. The stereotyped image of an elderly male patient with advanced-stage symptoms of PD can negatively affect patients' recognition of prodromal symptoms and their help-seeking behavior. It may also contribute to GPs' challenges in recognizing PD. GPs should apply a watchful waiting strategy with diagnostic safety netting to deal with the inevitable uncertainty that comes with the diagnostic pathway of PD. Moreover, GPs and patients should share their experienced feelings during this pathway. PD will then be diagnosed as soon as possible and the joint experience may strengthen the trusting patient-doctor relationship. This relationship is important to enable patients to express their care preferences and to offer support in formulating self-management goals that are not disease-oriented but focus on what is most meaningful to patients. Moreover, it facilitates shared decision-making. GPs' expertise in whole person medicine furthermore, allows them to coordinate all health care providers involved and to integrate care in a patient-centered manner by prioritizing problems in line with patients' preferences. Parkinson's disease in primary care should thus be a joint journey of community-dwelling patients and their GPs, in which a trusting relationship is of vital importance to offer much-needed personalized support.

The most important recommendations for clinical practice are the use of a watchful waiting strategy with safety netting during the diagnostic pathway and the need for awareness and confidence of GPs of the continuing added value of their care to community-dwelling PD patients. The focus for education should lie in creating a more representative image of the early presentation of PD, both for lay people and health care providers. Health care providers should also be taught the potential changes in care encountered by PD patients in order for them to be able to guide and educate their patients. Patients, in addition, should be taught to voice what is most meaningful to them and what support they need. Research should be performed in the way patients can best be educated in this, as merely inviting PD patients to share their needs and preferences may not be enough to overcome the difficulties they experience in communication and medical decision-making. As the general population is ageing and patients live at home for longer periods of time, the experiences and care needs of PD patients with advanced-stage disease and the way GPs can be supported to offer the care their PD patients need should also be the focus of research.

Samenvatting

Hoofdstuk 1 Algemene inleiding

In Hoofdstuk 1 beschrijven we de achtergrond, doelen en opbouw van dit proefschrift.
Al jaren voordat patiënten gediagnosticeerd worden met de ziekte van Parkinson kunnen ze klachten hebben, zogeheten prodromale symptomen. Deze symptomen zijn meestal niet ziektespecifiek. Voor patiënten kan het lastig zijn om te herkennen dat ze met hun klachten aandacht van zorgverleners nodig hebben en voor huisartsen is het lastig om te herkennen dat de aspecifieke klachten veroorzaakt worden door de ziekte van Parkinson. Als gevolg daarvan kan het traject tot de diagnose lang en onzeker zijn en negatieve gevolgen hebben voor de arts-patiënt relatie. Als de ziekte voortschrijdt treden er veranderingen op in de zorgbehoefte, veranderingen waar patiënten mee om moeten gaan. Ook al maken patiënten deze veranderingen van zorg misschien door zonder de betrokkenheid van professionele zorgverleners, zij kunnen nog altijd baat hebben bij ondersteuning. Huisartsen zijn ervaren in het bieden van zulke ondersteuning aan patiënten met veel voorkomende chronische aandoeningen.

Parkinsonpatiënten kunnen baat hebben bij vroegtijdige interventie en behandeling. Een vertrouwensband tussen arts en patiënt is essentieel om persoonsgerichte ondersteuning te kunnen bieden en gezamenlijke besluitvorming te faciliteren. Echter, kennis van de factoren die het verloop van het traject tot de diagnose en de beleving van patiënten tijdens dit traject beïnvloeden is gering. Inzicht in de prodromale symptomen die bij de huisarts gepresenteerd worden is bovendien beperkt: studies tot dusver hebben zich vooral gericht op klachten die bij de neuroloog worden gepresenteerd, klachten die vaak uitgesprokener zijn dan de prodromale symptomen die eerder in het ziektebeloop bij de huisarts gepresenteerd worden. Meer kennis van de ervaringen van parkinsonpatiënten met veranderingen in de zorg en de verwachtingen die zij hebben van de huisarts is bovendien nodig om persoonsgerichte ondersteuning te kunnen optimaliseren. Met dit proefschrift hopen wij meer inzicht te kunnen bieden in de ervaringen van patiënten voor, tijdens en na de diagnose ziekte van Parkinson en in de uitdagingen waar huisartsen mee worden geconfronteerd bij het bieden van zorg aan thuiswonende parkinsonpatiënten.

Op weg naar de diagnose ziekte van Parkinson

Het perspectief van de patiënt

Hoofdstuk 2 Tijdsintervallen in het diagnosticeren van de ziekte van Parkinson: het perspectief van de patiënt

Om meer inzicht te krijgen in de ervaringen van patiënten tijdens het traject van de eerste klacht tot de diagnose ziekte van Parkinson, hebben we een kwalitatieve studie uitgevoerd van essays over het diagnostische traject, geschreven door 902 patiëntleden van de Parkinson Vereniging (Hoofdstuk 2). Een gerichte steekproef van 52 essays is in een zich herhalend proces door twee onderzoekers bekeken, onafhankelijk van elkaar. De analyse is doorgegaan tot verzadiging was opgetreden. We vonden dat het traject tot de diagnose ziekte van Parkinson een zich herhalend proces is wat in drie tijdsintervallen kan worden

opgedeeld: herkenning van symptomen; het besluit om hulp te zoeken; en het proces van diagnosticeren. Patiënten beschreven dat het herkennen van de prodromale klachten van de ziekte van Parkinson lastig is aangezien het beeld dat zij van de ziekte hebben bestaat uit dat van bekende personen met een vergevorderd stadium van de ziekte van Parkinson. Als de klachten niet vanzelf overgaan of toenemen besluiten patiënten veelal professionele hulp te zoeken, een besluit wat beïnvloed wordt door factoren als de betrokkenheid van naasten en de houding van de patiënt tegenover gezondheidszorg. Patiënten beschreven daarnaast dat zij het gevoel hadden dat de huisarts ook moeite had om de ziekte van Parkinson te herkennen. We concluderen dat het noodzakelijk is om energie te steken in het uitbreiden van kennis over de prodromale symptomen van de ziekte van Parkinson bij leken en zorgverleners om het eerder stellen van de diagnose en vroegtijdig starten van de behandeling mogelijk te maken.

Hoofdstuk 3 Het traject tot de diagnose ziekte van Parkinson: een cross-sectionele survey studie naar factoren die van invloed zijn op patiënt ontevredenheid

In Hoofdstuk 3 presenteren wij de resultaten van de analyse van alle 902 essays van patiënten die we in Hoofdstuk 2 al noemden. Gebaseerd op de kwalitatieve analyse uit dat hoofdstuk, hebben we een methode ontwikkeld om de inhoud van de essays te coderen. Twee onderzoekers hebben onafhankelijk van elkaar meer dan 150 essays gecodeerd, initieel om tot consensus te komen over de codeermethode en later om dilemma's in het coderen te bediscussiëren. De andere essays zijn gecodeerd door één onderzoeker. De focus van dit onderzoek was de relatie tussen patiënt ontevredenheid en demografische gegevens, duur van het diagnostisch traject, communicatie met de huisarts en de neuroloog, het aantal betrokken zorgverleners buiten de huisarts en neuroloog, of een second opinion plaatsgevonden heeft en of er, volgens de patiënt, vertraging opgetreden is in het traject. Een subgroepanalyse is verricht om inzicht te krijgen in seksegerelateerde verschillen. Van alle patiënten was 16.4% ontevreden met het traject tot de diagnose en 4.8% zeer tevreden. De kans op ontevredenheid was groter bij patiënten met een laag opleidingsniveau, bij betrokkenheid van meer dan één zorgverlener, bij een second opinion op verzoek van de patiënt en bij vertraging veroorzaakt door zorgverleners. Als alleen de huisarts en neuroloog betrokken waren, waren vrouwen vaker ontevreden dan mannen. We concluderen dat als huisartsen zich bewust zijn van de risicofactoren voor ontevredenheid van patiënten en extra aandacht besteden aan communicatie en gezamenlijke besluitvorming, zij de ervaringen van patiënten positief kunnen beïnvloeden en bij kunnen dragen aan een vertrouwensband tussen arts en patiënt.

Het perspectief van de huisarts

Hoofdstuk 4 Prodromale symptomen en vroegtijdige herkenning van de ziekte van Parkinson in de huisartspraktijk: een nested case-control studie

De nested case-control studie die we presenteren in Hoofdstuk 4 is uitgevoerd om de prodromale klachten van de ziekte van Parkinson, die in de huisartsenpraktijk worden gepresenteerd, in kaart te brengen. Hiervoor hebben we gebruik gemaakt van de database van de Continue Morbiditeits Registratie (CMR). Deze database bevat informatie over

alle gezondheidsproblemen die ongeveer 12000 patiënten gepresenteerd hebben bij de huisartsen in vier praktijken. Uit deze populatie hebben we 86 parkinsonpatiënten en 78 gematchte controles geïncludeerd. We vonden dat parkinsonpatiënten in een periode van 2 jaar voorafgaand aan de diagnose ten opzichte van de controles vaker somatisch onvoldoende verklaarde lichamelijke klachten, obstipatie, hyperhidrosis en slaapproblemen presenteerden bij de huisarts. Patiënten hadden ook vaker last van meer dan één prodromale klacht en werden vaker verwezen naar eerstelijns zorgverleners of naar een medische specialist. We concluderen dat huisartsen alert zouden moeten zijn als patiënten meerdere symptomen presenteren over een periode van enkele jaren, meewegend wat het positieve effect zou kunnen zijn van vroege interventie.

Veranderingen in de zorg na de diagnose ziekte van Parkinson

Hoofdstuk 5 Veranderingen in de zorg bij de ziekte van Parkinson in de eerste lijn: protocol van een longitudinale mixed methods studie

Hoofdstuk 5 bevat het protocol van een longitudinale studie naar veranderingen in de zorg die thuiswonende parkinsonpatiënten meemaken. Het beschrijft werving, dataverzameling en data analyse in detail. De onderzoeksopzet was om een gerichte steekproef te nemen van huisartsen in en rond Nijmegen (Nederland) en hun thuiswonende parkinsonpatiënten zonder cognitieve problemen en hen voor een periode van 1 jaar te laten participeren in het onderzoek. Gedurende dat jaar zouden patiënten elke 14 dagen een videodagboek maken om verbaal en non-verbaal een indruk te geven van hun welzijn. Elke maand zou een onderzoeksassistent deze video's ophalen en informeren of er veranderingen in de zorg plaatsgevonden hadden. Als dat het geval was, zouden patiënten geïnterviewd worden over hun ervaringen en manier van omgaan met de verandering en over de rol die zij voor de huisarts zagen in de zorg voor parkinsonpatiënten. De informatie uit de video's zou hierbij gebruikt worden om de interviewgids aan te scherpen. Ook de huisartsen van de patiënten zouden geïnterviewd worden met de focus op hun rol tijdens de verandering in zorg die hun patiënt doorgemaakt had en op hun rol in de zorg voor parkinsonpatiënten in het algemeen. De geanonimiseerde transcripten van de interviews zouden worden geanalyseerd in een proces van continue vergelijking. Inclusie van huisartsen en patiënten, dataverzameling en data analyse zouden doorgaan totdat verzadiging was opgetreden.

Het perspectief van de patiënt

Hoofdstuk 6 Controle houden bij de ziekte van Parkinson: een kwalitatieve studie naar patiënt ervaringen

Hoofdstuk 6 beschrijft de ervaringen van thuiswonende parkinsonpatiënten met doorgemaakte veranderingen in de zorg en de manier waarop ze er mee omgegaan zijn. Hiervoor hebben we gebruik gemaakt van de interviews met 16 patiënten, die deelgenomen hebben aan de studie beschreven in Hoofdstuk 5. We vonden dat patiënten diverse veranderingen in de zorg meemaken zoals veranderingen in de ondersteuning van mantelzorgers, de aanschaf van hulpmiddelen, aanpassing van de medicatie of opname in het ziekenhuis. Drie thema's zijn van invloed op de ervaringen van patiënten en op hun acceptatie van eventueel resterende beperkingen: het anticiperen op verandering; zelfmanagement

van verandering; en het omgaan met verwachtingen. Patiënten kunnen zich overvallen voelen door een verandering in de zorg. Als ze erop kunnen anticiperen, draagt dit echter bij aan zelfmanagement van de verandering en aan het gevoel van controle. Patiënten gaven er de voorkeur aan de problemen die ze tegenkwamen zelf op te lossen. Zelfmanagement ging over het algemeen gepaard met realistische verwachtingen van de verandering en de acceptatie van resterende beperkingen, terwijl veranderingen die geïnitieerd werden door een zorgverlener gepaard gingen met onrealistisch hoge verwachtingen. We concluderen dat de veranderingen in de zorg die parkinsonpatiënten ervaren vaak nauw verbonden zijn aan hun persoonlijke situatie. Zelfs als ziektespecifieke zorg door specialisten wordt verleend, kan ondersteuning in zelfmanagement en het formuleren van realistische doelen hiervoor toch geboden worden door de huisarts die goed op de hoogte is van de faciliterende en belemmerende factoren in de situatie van de patiënt.

Het perspectief van de huisarts

Hoofdstuk 7 De ziekte van Parkinson: het perspectief van patiënt en huisarts op de rol van huisartsgeneeskunde

In Hoofdstuk 7 beschrijven we de rol die de huisarts volgens patiënten en huisartsen zou moeten hebben in de zorg voor parkinsonpatiënten. Om dit te kunnen doen hebben twee onderzoekers onafhankelijk van elkaar de eerder beschreven interviews met 16 thuiswonende parkinsonpatiënten en de interviews met 12 van hun huisartsen gecodeerd en gezamenlijk geanalyseerd volgens de principes van de continue vergelijking. We vonden dat patiënten de voorkeur gaven aan zelfmanagement van veranderingen en aan autonomie in besluitvorming. Om dit gedrag te stimuleren kozen de huisartsen ervoor een reactieve positie in te nemen bij patiënten met een nog niet vergevorderd stadium van de ziekte van Parkinson. Huisartsen voelden zich bovendien niet voldoende competent om hun rol uit te breiden. Patiënten hadden ook het idee dat de huisarts kennis en kunde tekort kwam en richtten zich met Parkinson gerelateerde vragen primair op de neuroloog. Huisartsen gaven daarnaast aan dat patiënten misschien niet goed zicht hebben op de rol die de huisarts zou kunnen hebben, een rol waarin huisartsen zelf het belang benadrukken van het bewaken van het welzijn van de patiënt. Hoewel de geïnterviewde patiënten geen extra rollen beschrijven voor de huisarts bij progressie van de ziekte, benoemen huisartsen een verschuiving naar een proactievere en uitgebreidere rol. We concluderen dat huisartsen meer vertrouwen zouden mogen hebben in de toegevoegde waarde van hun huisartsgeneeskundige benadering van de zorg voor patiënten met een complexe chronische aandoening zoals de ziekte van Parkinson. Parkinsonpatiënten halen er voordeel uit als huisartsen en specialisten elkaar aanvullen in de zorg die zij verlenen.

Hoofdstuk 8 Algemene discussie

In Hoofdstuk 8 plaatsen we de resultaten van de beschreven onderzoeken in perspectief. Bovendien bespreken we methodologische kwesties en geven we suggesties voor de dagelijkse praktijk, onderwijs en verder onderzoek.

We concluderen dat er vier thema's zijn die van belang zijn voor de zorg in alle fases van de

ziekte van Parkinson: het stereotype beeld dat er van de ziekte bestaat; de diagnostische onzekerheid waarmee de ziekte van Parkinson gepaard gaat; de toegevoegde waarde van generalisme aan de zorg voor parkinsonpatiënten; en het belang van een vertrouwensband tussen huisarts en patiënt. Het stereotype beeld dat van de ziekte van Parkinson bestaat, dat van de oude, voorovergebogen schuifelende man, kan een negatieve invloed hebben op de herkenning van de prodromale symptomen van de ziekte en op het hulpzoekgedrag van patiënten. Daarnaast kan het huisartsen bemoeilijken om de ziekte vroegtijdig te herkennen. Huisartsen zouden in het omgaan met de onzekerheid die inherent is aan het diagnostische traject van de ziekte van Parkinson een strategie moeten toepassen van 'watchful waiting', waarbij zij gelijktijdig zeker stellen dat patiënten terugkomen als dit nodig is. Zo kan de ziekte van Parkinson zo snel mogelijk worden gediagnosticeerd. Patiënten en huisartsen zouden bovendien hun gevoelens van onzekerheid moeten delen in deze fase aangezien dit kan bijdragen aan de vertrouwensband tussen arts en patiënt. Deze band is belangrijk om de drempel te verlagen voor het aangeven van eigen voorkeuren en prioriteiten door patiënten en om ondersteuning te kunnen bieden in het formuleren van doelen voor zelfmanagement die niet ziektegericht zijn maar zich richten op wat de patiënt het belangrijkst vindt. Bovendien faciliteert een vertrouwensband tussen arts en patiënt gezamenlijke besluitvorming. De expertise van huisartsen in het bieden van 'whole person medicine' leidt er bovendien toe dat zij de zorg zo kunnen coördineren en integreren dat die gebaseerd is op de voorkeuren en prioriteiten die de patiënt aangegeven heeft. Thuiswonende parkinsonpatiënten en hun huisartsen zouden de reis, die de ziekte van Parkinson is, dan ook gezamenlijk moeten afleggen, waarbij een vertrouwensband essentieel is om persoonsgerichte ondersteuning mogelijk te maken.

De belangrijkste aanbevelingen voor de huisartsenpraktijk zijn het gebruik van een 'watchful waiting' strategie gedurende het diagnostische traject van de ziekte van Parkinson en het herkennen van en vertrouwen hebben in de toegevoegde waarde van generalistische zorg voor thuiswonende parkinsonpatiënten. De focus voor onderwijs zou moeten liggen in het tot stand brengen van een representatiever beeld van de prodromale fase van de ziekte, zowel bij leken als bij zorgverleners. Zorgverleners zouden bovendien meer kennis moeten krijgen van de mogelijke veranderingen in zorg die parkinsonpatiënten kunnen ervaren, zodat patiënten hierover beter geïnformeerd en hierin beter begeleid kunnen worden. Patiënten zouden geleerd moeten krijgen om uit te spreken wat zij belangrijk vinden en welke ondersteuning zij nodig hebben. Onderzoek zou gedaan moeten worden naar de manier waarop zij dit het beste zouden kunnen leren, aangezien parkinsonpatiënten enkel uitnodigen om hun behoeften en voorkeuren te delen mogelijk niet genoeg is om de moeilijkheden die zij ervaren in communicatie en medische besluitvorming te overwinnen. Met de vergrijzing van de bevolking en het langer thuis wonen van hulpbehoevende ouderen zou eveneens onderzoek gedaan moeten worden naar de ervaringen en zorgbehoeften van patiënten met een verder gevorderd stadium van de ziekte van Parkinson en naar de manier waarop huisartsen ondersteund kunnen worden in het bieden van passende zorg aan hun parkinsonpatiënten.

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Ik hou van jullie!

Curriculum Vitae

Annette Plouvier werd geboren op 17 juli 1981 in Uden. Ze behaalde in 1999 haar gymnasiumdiploma aan het Udens College in Uden, waarna ze geneeskunde ging studeren aan de Radboud Universiteit in Nijmegen. In 2005 rondde ze deze studie af. Vanwege haar voorkeur voor een persoonsgerichte benadering van zorg en haar interesse in complexe problematiek, revalidatie en palliatieve zorg ging zij werken als anios ouderengeneeskunde bij Pantein in Boxmeer. Tussen 2006 en 2008 volgde zij hier ook de opleiding tot specialist ouderengeneeskunde, een functie die zij vervolgens korte tijd bij BrabantZorg in Oss bekleedde.

De theoretische achtergrond bleek echter beter bij Annette te passen dan de praktijk. Tussen 2009 en 2011 werkte zij als regiocoördinator bij het Bijwerkingencentrum Lareb. Toen zich in 2011 de kans voordeed om 'op het oude nest' - het Radboudumc - onderzoek te gaan doen bij de afdeling Anesthesiologie, Pijn en Palliatieve Geneeskunde twijfelde zij geen moment. Hiermee had Annette de smaak te pakken en na afronding van het onderzoeksproject in de palliatieve zorg startte zij in september 2013 met haar promotieonderzoek 'Parkinson's disease in primary care, a joint journey of patients and general practitioners'. Annette zal na haar promotie als postdoc blijven werken op de afdeling Eerstelijnsgeneeskunde van het Radboudumc.

Annette woont samen met Jouke Landman en hun drie kinderen Janne (2010), Siem (2012) en Jet (2015) in Nijmegen.